Developmental Coordination Disorder (DCD) as a Distinct Syndrome: A Conceptual and Empirical Investigation

Judith Mary Peters

A thesis presented in partial fulfilment of the requirements for the degree of Doctor of Philosophy in Psychology

Institute of Education
University of London

2006

Abstract

Bright children who have abnormal difficulty in acquiring age-appropriate motor skills are of increasing concern to parents, teachers and health professionals. Longitudinal studies have found the condition to be associated with educational under-achievement, impaired social development and disturbed mental well-being in adult life. Now officially known as Developmental Coordination Disorder (DCD), the question of whether this condition should really be viewed as a distinct diagnostic entity is the central theme of this thesis.

In the opening chapters, the history of terms used to signify 'clumsiness' of movement is reviewed and the different implications of treating such behaviour as a symptom or syndrome is considered. Discussion then moves to the overlap between DCD and other childhood conditions and the question of how these should be conceptualised.

Five studies comprising the empirical component of the thesis employed a variety of methodologies. Two questionnaire-based studies showed that in this area, neither consensus on terms nor equitable service provision has yet been achieved. A third, retrospective study, searched for evidence of sub-types within a large sample of DCD children, successfully replicating some of the cluster groups reported by others. In a final, prospective study, a two-stage identification process was followed by 'blind' assessment of boys with DCD, Asperger Syndrome or Joint Hyper-mobility Syndrome. Novel to this area was the inclusion of experimental measures, including dual-task performance, in which motor and cognitive tasks were combined. The results showed that although the group with AS were significantly poorer on ball skills than those with DCD, the general nature of motor difficulties was not systematically constrained by diagnosis. Together, these studies support the thesis that DCD exists as a separable syndrome, but bear less decisively on the existence of subtypes. A series of real-life case studies illustrates the problems associated with differential diagnosis and the implications for appropriate intervention.

Declaration and Word Count

All the empirical studies reported in this thesis were planned, executed and analysed by the candidate. Co-authors in Studies One and Two participated in the discussion results and commented on the write up of papers for publication. The candidate also confirms that the work submitted is her own and that appropriate credit has been given where reference has been made to the work of others.

Word count: 94951 words

Judith M. Peters

Acknowledgements

This research was supervised by Sheila Henderson. I am deeply grateful to her for her time, energy, perfectionist demands, patient supervision and positive encouragement throughout.

I am grateful to the Physiotherapy Department at GOSH who accommodated the outpatient assessments, and staff who gave support and encouragement (Head of Department, Devala Dookun; Acting Head, Rosa Schmale). I thank Beverleigh Christian, Nathan Hasson, Betty Hutchon, Alice Jones, Sue Maillard, and Angela Webb for referrals to the final research study and am particularly indebted to Rebecca Worsley and Monica Murray for co-ordinating the initial referrals and administering the vocabulary test, assisted by Debbie Jackson.

Many others have provided advice for this work. In particular the following deserve thanks: Peter Hill for initial fruitful discussions of the project, provision of assessment tools and links to referral sources; Charlie Owen for patient advice on statistics; Eric Hadley for constructing the peg board for Study 5; Monica Booth and Gwen Dornan for assessing handwriting samples; Dan Shaw for attending to frequent computer idiosyncrasies; Kate Sym for assistance with formatting the final manuscript; Ellen Cotter for her assistance, Leslie Henderson for his perspicacious comments and Cordon Bleu expertise.

I am grateful to Lucy Alderson, Anna Barnett, Melanie Bladen, Eleanor Main, Liz Stephenson and many others for helpful discussions, friendship, support and encouragement.

I acknowledge the help of schools who referred children but above all I am indebted to all the parents and children who took part in the research. From them I learnt much about the real impact of movement competence in every day life.

Finally, I thank my children, Carolyne, Sarah and Christopher for their moral support and encouragement throughout an exciting and challenging journey. Huge thanks also go to my husband, Timothy who tolerated my absorption in this research and who read many drafts of the manuscript with a critical eye, and provided essential support at the times when it mattered most.

List of Abbreviations

ADD/ADHD	Attention Deficit Disorder/Attention Deficit Hyperactivity Disorder
AHP	Allied Health Professional
AS	Asperger Syndrome
ASD	Autism Spectrum Disorder
ASSQ	High Functioning Autism Spectrum Screening Questionnaire
BJHS	Benign Joint Hypermobility Syndrome
BPVS	British Picture Vocabulary Scales
CD	Conduct Disorder
CP	Cerebral Palsy
DAMP	Disorders of Attention, Motor Control and Perception
DCD	Developmental Coordination Disorder
DMD	Duchenne Muscular Dystrophy
DSM	Diagnostic and Statistical Manual
FTND	Full Term Normal Delivery
GOSH	Great Ormond Street Childrens' Hospital
HFA	High Function Autism
ICD	International Classification of Diseases
LSCS	Lower Section Caesarean Section
M-ABC	Movement Assessment Battery for Children
MBD	Minimal Brain Dysfunction
MD-hyp	Motor difficulty plus hypermobility
MD-nonhyp	Motor difficulty without hypermobility
NF1	Neurofibromatosis Type 1
NVLD	Non Verbal Learning Disability
OCD	Obsessive Compulsive Disorder
ODD	Oppositional Defiant Disorder
OED	Oxford English Dictionary
OT	Occupational Therapist/therapy
PDD	Pervasive Developmental Disorder
PDD-NOS	Pervasive Developmental Disorder Not Otherwise Specified
PIQ	Performance IQ
PT	Physiotherapist/therapy
RD	Reading Disorder
SALT	Speech and Language Therapist
SDD-MF	Specific Developmental Disorder of Motor Function
SDQ	Strengths and Difficulties Questionnaire
SENCO	Special Education Needs Coordinator
SIPT	Sensory Integration and Praxis Tests
SLI	Specific Language Impairment
TD	Typically Developing
TS	Tourette Syndrome
VIQ	Verbal IQ
VMI	Developmental Test of Visual Motor Integration
V-P	Verbal-Performance
WISC	Wechsler Intelligence Scale for Children

Contents

List of Tables

List of Figures

Chapter 1

"Clumsiness" of Movement: A Historical Review

1.0 Introduction

Leonardo Da Vinci wrote that movement is the cause of all life (MacCurdy, 1938). It is a fundamental characteristic of all living things. Motor behaviour provides the means by which an organism interacts with its environment and as such is of major biological and social importance. Most adults have at their disposal a full repertoire of basic movement skills, which they use so automatically, and with such ease, that smooth co-ordinated action is produced without any conscious thought. They have acquired an essential foundation for adaptive function as sociable human beings.

In contrast to the full grown adult, the human baby begins life as a motorically helpless creature, entirely dependent on the adult for survival. The process of development from helplessness to competence is a long and complex one, involving many different systems in constant interaction with each other. In order to survive and prosper the mammalian brain must become tuned or programmed to the environment via perception and motor action and interaction. The human machine is comprised of more than two hundred bones, over three times as many individual muscles connected via ligaments tendons and movable joints. Muscle and joint flexibility creates, in mechanical terms, degrees of freedom, defined by Kelso (1982, p. 23) as "potential variables that must be controlled if we are to function efficiently". Control of so many degrees of freedom requires a tripartite neural network: nerve impulses arising in the brain and spinal cord (central nervous system), transmitted to and from body structures through a network of nerves (peripheral nervous system) and underpinned by simultaneous regulation and co-ordination of vital body structures by the autonomic nervous system.

In the past, the debate about how the process of development might be conceptualised took a polarised form between, on the one side a predetermined genetic model of development (nature dominates) and on the other, one constructed through environmental factors (nurture dominates). Nowadays, such polarisation would be considered too simplistic. Recent evidence supports interaction between genetic and environmental influences throughout the life span. Plasticity is

considered to be a fundamental property of early brain development (Johnson, 1997). Plasticity refers to flexibility or ability to undergo change. Plasticity is also recognised in adaptation of skeletal muscle in response to function (Pette & Vrbova, 1995). Thus plasticity occurs and allows scope for change to occur within both perceptual and motor systems. Research in neurodevelopment increasingly moves forward from a static hierarchical conceptual model toward an intricate picture of inter-dependent systems and subsystems switched on and off at critical times by various triggers including neurotransmitters. The development of the nervous system is influenced by the interaction of both genetic and environmental factors. The complex process of development involves orchestrated growth of new cells, changes in cell location (migration) and selective programmed cell death (apoptosis). Cell specificity is induced and tissues differentiated for function. The processes, which dictate the formation and remodelling of dendrite and synapse connections, emanate both from within the organism (centrally) and are also peripherally driven by behavioural, perceptual and motor influences (Brown, Tarek, & O'Regan, 1997; Johnson, 1997).

Muscle action is involved throughout the human body systems - musculo-skeletal, respiratory, digestive, circulatory, neurological, endocrine, immune and renal-reproductive. In an adult some 40% of the body mass is muscle and as the largest organ in the body is central to nutrition and fluid and electrolyte metabolism (Edwards, 2002, p. xvii). Fuelled by glucose and fatty acids from nutrients, and oxygen from blood, muscles convert chemical energy into force and mechanical work. Muscles play a continuous role in cardio-respiratory functions and the stabilisation of posture. They power all actions from the precision required by hands and eyes for threading a needle to the larger body movements needed to climb a mountain. Two distinct types of muscle fibres, red and white further define specific functions, which can adapt and change according to need. The former, mitochondria-rich red fibres are responsible for sustained action (e.g., marathon running), whereas white fibres are highly efficient for short bursts of muscle action (e.g., sprinters).

1.1 Motor dysfunction

In view of the complexity of human sensory-motor control it is little wonder that disruption in any of the systems of the body can lead to movement dysfunction. In childhood, for instance, disease/impairment of the body's framework may

compromise movement. Bones (e.g., brittle bones, limb deformities), joints (arthritis) tendons/ligaments (collagen diseases, hypermobility) may be affected and jeopardise everyday function at home and in school. To give one example, childhood arthritis which affects a few or very many small or large joints occurs, often with insidious onset, in children of any age.

Dysfunction may result from muscle abnormalities in either microscopic or macroscopic terms - from smooth muscle within arterial walls, cardiac muscle (which pumps blood throughout life), to skeletal muscles, which are under voluntary control. For instance, muscular dystrophy results in progressive muscle weakness. Neurological pathology also results in movement dysfunction via the central nervous system (e.g., cerebral palsy, corpus callosum agenesis), peripheral nervous system (e.g., neuropathies), and sensory systems (visual impairment, auditory/vestibular damage). Additionally movement can be affected by disorders of metabolism, (metabolic myopathies), endocrine dysfunction (diabetes mellitus) and nutritional deficiencies (protein, vitamin deficiencies). Not only can the impairment be classified by system but also by the timing (congenital, acquired, degenerative, temporary) and by aetiology: infections (meningitis, poliomyelitis); toxins (lead, alcohol, drugs); genetic (Down, Ehlers Danlos, Fragile X). Taken together, these various system failures lead to a myriad of known medical conditions which affect movement (see Dewey & Tupper, 2004).

Over the last century reports have been published in the literature, of children who do not suffer from any known disease and do not fit neatly into any of the diagnostic categories referred to above yet lack the motor competence to cope with everyday living. For example, children who were "clumsy" were mentioned by the French paediatrician Dupré as early as 1909 (Dupré & Merklen, 1909) and described in more detail by Orton (1937, p. 121), who provides the now classic description: "Such children are often somewhat delayed in learning even the simpler movements such as walking and running, and have great difficulty in learning to use their hands and to copy motions shown to them. They are slow in learning to dress themselves and are clumsy in their attempts to button their clothes, tie their shoes, handle a spoon, and in other simple tasks". Seventy years on these key descriptors can be found in the formal diagnostic manuals published by the World Health Organisation (WHO, 1992) and American Psychiatric Association (APA, 2000), discussed later in this

chapter. For example, the ICD 10 (WHO, 1992) description includes the following "...slow to learn to hop, run and go up and down stairs...difficulty tying shoe laces, to fasten and unfasten buttons, and to throw and catch balls. The child may be generally clumsy in fine and/or gross movements - tending to drop things, to stumble, to bump into obstacles, and to have poor handwriting" (p. 251).

Many terms have been used to describe children with these characteristics and there is continuous debate about how to conceptualise the difficulties they encounter. There are a number of reasons why there has been such confusion, some of which will be discussed in the first three chapters of this thesis. In the present chapter, the primary focus of attention will be the problems caused by the use of different terminology. In this context, the view taken is that the movement difficulties these children experience can be conceptualised as a single discrete entity or syndrome. In Chapters 2 and 3 of the thesis, however, this view is qualified and two other questions are posed. In Chapter 2, attention shifts to the question of whether the syndrome is in fact unitary, or alternatively consists of a number of clinically meaningful sub-types. Then in Chapter 3, the wheel turns full circle to the question of whether movement difficulties of concern here are simply one component of a broader mix of other developmental difficulties.

1.2 Terminology: sources of confusion

Although several authors have discussed the confusion caused by the application of so many different labels to children with movement difficulties (e.g., Aicardi, 1992; Henderson & Barnett, 1998; Missiuna & Polatajko, 1995) little attention has so far been placed on the original meanings of the words, the history of their use over time, or the problems associated with specific "professional languages". In what follows, the chaos surrounding the labelling issue is discussed under four headings:

- The use of lay terms as labels for "medical" conditions.
- The application of terms derived from adult neurology to childhood conditions.
- The idea of a continuum of brain damage in relation to movement disorders.
- Geographical and professional individuality.

1.3 The use of a lay term as a label for a "medical" condition

There is little doubt that one of the worst sources of confusion surrounding the definition of the condition of concern here is the use of the term "clumsy". The crux

of the matter is that the adjective "clumsy" has been used both as a descriptor of the motor dysfunction that we are concerned with, and as the label for the medical condition which has the motor dysfunction as its defining feature. To a lesser extent, the words physically awkward have been treated in the same way.

1.3.1 Clumsiness as a "normal" phenomenon

Temporary or intermittent, motor difficulty is familiar to everyone at some time or another and a plethora of descriptive words exists in common English usage to describe such behaviour. Examples include "awkward", "clumsy", "gawky", "blundering", "bumbling", "ungainly", "unhandy" "uncoordinated", and "unskilful". Similarly, many, often colourful, idioms exist which not only describe the behaviour in question but also add insights into the cause of the lack of dexterity. For instance the phrase "His fingers are all thumbs" implies that thumbs are not as flexible instruments of movement as fingers.

The clumsiness that we all consider to be "normal" and characteristic of all of us throughout the life span takes different forms and requires different kinds of explanation. The following examples illustrate how varied these explanations are - and by implication, show how difficult it can be to separate "normality" from "abnormality".

"Natural" clumsiness as part of normal stages of maturation

Natural clumsiness can be observed throughout the life span. It is obvious in extreme youth (e.g., the baby's early attempts to grasp, the toddler's unsteady gait), and in very old age (e.g., fumbling with coins, insecure balance). What makes things difficult is trying to decide what is "normal" and where abnormality creeps in. The three-year-old, who carefully carries an egg but accidentally breaks the shell, demonstrates a lack of ability to grade grip strength – quite normal at this age and not indicative of clumsiness in a medical sense. Conversely, the three-year-old who cannot grasp and pick up a cup or beaker for a drink is probably "abnormal".

The effect of physical constraints on movement

Simple mechanical constraints can lead to clumsy movement in all of us. For example when over-weight during pregnancy, simple locomotion looks awkward and ungainly and is often exhausting - but is nevertheless a "normal" feature of changing

body shape. Attempts to function when out of proportion to the surrounding space also results in "normal" clumsiness' for many of us. Expert watchmakers and surgeons may be able to manipulate minute objects with ease but most of us struggle. The skilled furniture remover may carry large furniture in a confined area with an impressive ease that few of us can emulate.

The effect of other challenges from the internal or external environment

Challenges to all of the senses may lead to clumsiness of movement at some point in time. For example, when the tactile senses are challenged by a slippery surface, postural control becomes difficult and a perfectly normal fall may occur. Clumsiness is also noticeable when the body is very cold, tired, hungry, or sick. We can all remember an occasion when our hands were so cold that it became impossible to carry out fine movements, or the experience of staggering dizzily from a merry-go-round. Rather similar motor difficulty arises when the body biochemistry is altered by substances such as alcohol, or when the brain is starved of oxygen e.g., at high altitude.

Stress

Stresses of various kinds may compromise movement performance. For example a task may prove too demanding through attempts to perform an action too fast and a point will eventually occur when motor control breaks down and skills disintegrate; "The more haste the less speed" (Heywood, 1546 cited by Hyman, 1989). Stress is revealed by the all too familiar experience of trembling hands that accompany anxiety, anger or fear: "shaking like a leaf"; "frozen with fear". "Shaking" or "freezing fearfully" may be seen as a normal response but how much more magnified that response can become in clumsy children when stressed, with obvious repercussions on such functions as handwriting.

1.3.2 Clumsiness as an abnormal phenomenon

According to Orton (1937, p. 120) the idea that some children exhibit a degree of clumsiness that could be viewed as *abnormal* goes back at least to Galen (131-200 AD). As surgeon to the school of gladiators, Galen devoted himself to medicine and was renowned for his pioneering description of the muscular system (Singer, 1928). Galen is reported to have spoken of some children as being "ambilevous", or "doubly left-handed". Galen wrote in Greek but his work was generally accessed through

Latin translation hence "ambilevous" (Nutton, 2005). Although Orton alludes to this historically interesting "medical" reference to children who lack dexterity, the idea that clumsiness of movement might be linked to some sort of medical condition is a relatively recent notion when compared to the use of the word in everyday language.

Over the years, the term "clumsy" has consistently been the commonest descriptive term used in both lay and professional language to describe children whose clumsiness is considered to be abnormal (Missiuna & Polatajko, 1995). The term clumsy appears in the titles of publications in medical, paramedical, educational, and psychological literature as well as in everyday language. A cursory search of the literature found at least one reference published every year since 1961 (e.g., Barnett & Henderson, 1992; Gordon & McKinley, 1980; Gubbay, 1975; Hadders-Algra & Gramsbergen, 2003; Walton, 1961). Because of its derogatory connotation, however, there has been a gradual trend toward avoidance of the use of the term and by 2006 it is certainly deemed "politically incorrect" in professional circles. The term "awkward" has been used less frequently than "clumsy" but was used by Keogh (1968) and also preferred by Wall, who has had a strong influence over the direction of the present Canadian conceptualisation of terms (Marchiori, Wall, & Bedingfield, 1987; Wall, Reid, & Paton, 1990). Whether it was a good idea to apply the exact same terms to denote behaviour that is actually outside the understanding of most "men and women in the street" is the crux of the matter.

1.3.3 An etymological search

Although it might not have been a good idea to use the same term to describe behaviour that is both normal and abnormal, this is in fact what has happened. In order to understand better why such confusion arose an examination of the origins of the words clumsy and awkward might well be fruitful. Clearly, the starting place for such a quest is a dictionary, which traces the etymology and historical use of a particular word. The Compact Oxford English Dictionary (OED, 1991), first published between 1884 and 1928, has established itself as one of the best sources of reference. In lexicographical terms, it is a diachronic dictionary; that is, it not only defines the language of the present day, it also records its use at any period within the Dictionary's coverage (i.e., words used since the 12th century excluding words which were obsolete by the year 1150). In addition to explaining what a word form means, it also, by documenting its history explains why it has come to have a particular

meaning. Virtually any source of the printed word is used to illustrate, through quotations, a comprehensive history of the English language (OED, 1991, p. 3). Sources both scholarly and popular include journals, magazines, newspapers, Biblical text and government documents, manuscripts, collections of letters and diaries.

A search of the OED reveals that the terms "clumsy", and "awkward" are often used synonymously, share early origins and were both used before 1600 (see Table 1.1). The OED quote by Swift "I have not seen a more clumsy, awkward, and unhandy people" (see Table 1.1) suggests that the words, "clumsy", "unhandy" and "awkward" are all synonyms. This entry conveys an interpretation of the word "awkward", which reflects deviation from the norm and the idea of ridicule is brought in by Shakespeare in Troilus and Cressida. The definition involves not only a reference to direction/laterality but also the actions of a person (subject) and the part played by an object (use of tools). Additionally behaviour is described as "perverse in nature or disposition" suggesting both trait and state.

The quotations from the OED also provide interesting links to current research into the causes of abnormal clumsiness in children. For example, words and phrases such as: "benumbed", "frozen", "bereft of sensation", and "lack of power – palsied", "power of grasping", "heavy" suggest both sensory dysfunction and loss of motor power. Reference to "lock-jaw/speechless" having a common root is interesting as Orton (1937) linked speech and language problems to clumsiness in his study of several hundred children with language delays and disorders. He cites Sir Richard Paget's thesis "that the sign language was the first form of symbolic language to be developed and that spoken language was largely an outgrowth therefrom" (ibid, p. 17). Although mention of 'lock-jaw' is not lexicographic the link between communication and movement especially in relation to symbolic gesture becomes a recurrent theme in many studies involving children with developmental delays (Duel & Doar, 1992; Hill, Bishop, & Nimmo-Smith, 1998; Miller, 1988; Powell & Bishop, 1992; Square, Roy, & Martin, 1997).

Table 1.1 *Clumsy and Awkward, Origin Earliest Quotes and References (OED, 1991)*

Clumsy 1600

The word is used by writers before about 1600 but was not used by Shakespeare. It is derived from clumse + y The localisation of the word in England agrees with a Norse origin. The obsolete meanings given:

1. Benumbed or stiffened with cold.
   1600 "The Carthaginians...returned into the campe so clumsie and frozen" (Holland. Livy xxi lvi. 425)
2. Acting or moving as if benumbed: heavy and awkward in motion or action; ungainly, unhandy; wanting in dexterity or grace.
   1597 -8 "When each base clowne his clumsie fist doth bruise" Bishop Hall Sat.1.iii.42)
3. Applied to actions and products of clumsy hands: ill-contrived, awkward.
   Clumsed or clumst - benumbed with cold; numb, palsied, bereft of sensation and power of grasping. Similar words appear in modern Scandinavian - Icelandic, Klumsa = lock-jawed, speechless.

Awkward 1530 –

The word probably stems from old Norse awk+ward i.e., "in an awk direction".
Awk (Obsolete) is given three meanings:

1. Directed the other way or in the wrong direction, back-handed, from the left hand.
2. Untoward, froward, perverse, in nature or disposition.
3. Untoward to deal with, awkward to use, clumsy.

Therefore the original meaning given for awkward was "in the wrong direction, in the wrong way, Upside down; hindside foremost; In a backward direction, with a back stroke; asquint".

1. "Awkwar, leftehanded, gauche" (1530)
2. "I have not seen a more clumsy, awkward, and unhandy people" Swift Gulliver (1816) III ii 189.
3. "With ridiculous and aukward action ....he pageants us" (1606) Wm Shakespeare. Troilus & Cressida. iii 149.

"A *variety of awkward gambols*" (1865) Dickens Charles Mut. Fr. vii 314.

"Clumsiness" is a world-wide phenomenon and the word "clumsy" and its synonyms found in the English language are also included in dictionaries in other languages. Table 1.2 lists translations given for "clumsy" from a selection of current dictionaries. It is interesting to note how many of these terms have an onomatopoeic ring to them e.g., Lomp (Dutch), Plump (German).

Table 1.2 *"Clumsiness" in Various Languages (Adapted from Burnham, 2005)*

Language	Word	Implied meaning
Dutch	Houterig, kluns	Clumsy
	Onhandig	Clumsy, awkward, flat-footed
	Lomp	Ungainly
	Plomp	Clumsy
Finnish	Kömpelo	Lacking in motor skill
French	Maladroit	Clumsy (person, physically), awkward, unskilful,
	Gauche	Clumsy (person figuratively), stupid, foolish, tactless
Greek	Atsalos ατσαλοσ	Clumsy
	Adexio αδεξιο	Not dextrous
German	Unbeholfen	Clumsy, awkward, bungling, fumbling, ungainly, brusque, blunt, bluff
	Schwerfällig	Dull, slow, clumsy
	Ungeschickt	Awkward, clumsy, unskilful, gauche, inept, maladroit
	Plump	Podgy, clumsy, awkward, shapeless, heavy, crude, ill-bred, tactless, blunt
Hungarian	Ügyetlen	Maladroit, unhandy
	Kètbalkezes	Negative: a person with two left hands
	Béng	Slang: a paralytic person
Icelandic	Klaufalegur:	Clumsy
Italian	Maldestro	Awkward, clumsy
	Malaccorto	Incautious, imprudent, rash, awkward
	Goffo	Clumsy, awkward, stupid
Portuguese	Desajeitado	Awkward, clumsy, unskilful, uncouth
	Tosco	Coarse, rough, crude, clumsy, awkward
Spanish	Torpe	Clumsy, dull-witted, stupid
	Patoso	Clumsy-footed; unintentionally funny
	Desmañdo	Clumsy, awkward
Welsh	Clogyrnaidd or	Clumsy, awkward, rough
	Lletchwith	Clumsy, awkward
Yiddish	Klotz or Klots	Ungraceful, awkward, clumsy person; bungler (Eng. slang "klutz")

What this brief historical review has shown is that the term "clumsy" and its various synonyms have commonly been used as descriptive terms worldwide for hundreds of years. We all know what the terms mean and we all have some idea of the kind of behaviour being described.

1.3.4 Attitudes to "clumsiness"

The notion that lack of coordination is something to be made fun of and mocked is common in all societies. The circus clown in baggy trousers and oversize shoes is a caricature of the concept of clumsiness: ridiculed by all and associated with stupidity. Ridicule and accident proneness is suggested in "each base clowne his clumsie fist

doth bruise" (see Table 1.1). Such negative views are expressed verbally when a person is described as a "butterfingers" or "a bull in a china shop". Less directly, the phrase "the greatest talkers are (always) the least doers" (16th C cited by Hyman, 1989) implies that talking and doing are not always well coupled. Although many of these terms and phrases are often used without malice, there is a thin line between what may be perceived as harmless teasing by the donor and hurtful criticism by the receiver. Moreover, the use of such language can enter the realm of bullying and intimidation for the child who is "abnormally" clumsy.

The idea that clumsiness might be avoidable is one that can be found in idioms that also have a long history. For example, "Festina Lente" (literally "haste slowly") was reportedly the motto of Emperor Augustus (31BC-41AD). Similarly "look before you leap" (14th C cited by Hyman, 1989); "put one's best foot forward"; "slow but sure" (Clarke, 1639 cited by Hyman, 1989); and "practice makes perfect" (Latin, cited by Hyman, 1989) all reflect the layman's intuitive understanding of the need for planning one's action ahead of time. Although practice **may** lead toward perfection, however, practising the wrong action will undoubtedly be counter-productive, a view that is not always understood by professionals dealing with children with abnormal clumsiness.

Another belief that has been around for some time is that left-handedness is inevitably associated with "clumsy" movement. An unusual term with Latin origin, is ambilaevous or ambilevous. This term is cited (OED, 1991) as in use in 1646 "ambilevous or left handed on both sides" and a further example given for 1878 "Having left hands only; that is clumsy". Maligning clumsiness as somehow "abnormal" and to be scorned or scoffed at derives from common views of left-handedness per se. Worldwide, approximately 90% of people are right-hand dominant. The comparative paucity of left-handers has led to cultural stigma and to a sense of abnormality and peculiarity reflected in derogatory terms derived from words such as "sinister" and "gauche". Many expressions infer disapproval of left-hand dominance: "keggy fisted"; "cack-handed"; "southpaw". In contrast, the positive advantages of sinistrality are rarely mentioned e.g., in sports such as tennis, cricket, boxing or the benefits of ambidexterity for some musicians e.g., percussionists.

In many instances, "natural" clumsiness may be ridiculed but is generally perceived as little more than a nuisance. When the end result is harm either to the individual or to others, however, a different emphasis and importance is implied encompassing blame, carelessness and inefficiency. Perhaps not surprisingly one of the places where clumsiness may not be accepted as harmless is in the workplace, where it can lead to damaging outcomes. What is surprising, however, is that there is a substantial literature on the causes of accidents in workplaces and their avoidance, which refers to clumsiness but is almost never accessed or referred to by those professionals interested in "clumsy" children. Perhaps one reason for this is that accident-proneness clearly does overlap with clumsiness but is not synonymous.

Although accidents were no doubt a source of concern, much of the work related to accidents originated with the rise of machinery during and since the Industrial Revolution and emerged alongside both, protection of the individual and the management, through health and safety and insurance protocols. The concept of a human factor contributing to accident occurrence was first mentioned in the early 20th century (Editorial, 1919) and Burnham (2005) views the term "clumsiness" as a forerunner from the perspective of susceptibility to accidents to the idea that some individuals might be "accident prone" (Burnham, 2005).

1.3.5 Clumsiness as a syndrome

In the UK, Walton was one of the first and most influential people to refer directly to a discreet "clumsy child syndrome" (Walton, 1961; 1963; Walton, Ellis, & Court, 1962). By attaching the medical term "syndrome" to the label "clumsy", Walton in effect formally drew the common English word "clumsy" into medical vocabulary and thus was responsible for much of the confusion encountered in the next 30 years.

An editorial, entitled *Clumsy Children* published in the British Medical Journal (BMJ Editorial, 1962) draws the attention of doctors to this pervasive and insufficiently researched topic. It cites the work of Annell (1949) and describes the clumsy child as "awkward in his movement, poor at games, hopeless in dancing and gymnastics, a bad writer, and defective in concentration. He is inattentive, cannot sit still, leaves his shoelaces untied, does his buttons wrongly, bumps into furniture, breaks glassware, slips off his chair, kicks his legs against the desk, and perhaps reads badly" (BMJ Editorial, 1962 , p1665) Unfortunately, Annell of Uppsala, Sweden chose the term

"motor (motorial) infantilism" to describe motor dysfunction occurring in children with normal intelligence, thus classifying the problem as largely one of delayed maturation. Her suggestion of a good prognosis is perhaps what led to so many parents being told "Don't worry he will grow out of it".

The British Medical Journal editorial cites three other reports pertaining to childhood clumsiness, which influenced later thinking. These studies, (Illingworth, 1963; Prechtl & Stemmer, 1962; Walton et al., 1962) contrasted with that of Annell, in that longer term and more widespread effects in movement, concentration and school achievement were emphasised. It is noteworthy that at this initial emergence of a clumsy "syndrome" several concurrent studies are cited which agree on the general features of clumsiness, while at the same time, emphasising and highlighting different symptoms. These disparities are discussed in more detail in the next chapter.

Since "clumsiness" became established as a medical syndrome, the term "clumsy" has continued to be used **both** as a descriptor and as a label, particularly in the UK. Grimley and McKinley (1977, p. 3) comment "clumsy is a qualitative adjective like "stylish" or "beautiful". It is used as the result of a general impression of an individual's performance of a range of activities and is not complimentary!" This comment reflects both the historical links with "clumsy" as a derogatory term mentioned earlier, and responses made in the first study reported in this thesis.

At the same time as the word "clumsy" was being used as a label for a distinct clinical entity/childhood disorder other terms were being used as labels for children who seemed, on the face of it, very similar to those in the above discussion. These include the term "dyspraxia" and its variants and the term "minimal brain damage" and its variants. Each of these will be discussed below.

1.4 Dyspraxia and its variants: Terms derived from adult neurology applied to developmental childhood conditions

In 1960 Polani commented "Paediatric neurology as a speciality does not exist in Britain today...", whereas "...neurology of adults is a well established, solid, ancient and especially distinguished speciality" (Polani, 1960, p. 5). This statement helps us understand why it is not uncommon to find a childhood condition bearing a label

derived from adult neurology. This has led to misunderstanding and much confusion, mainly because the developmental version of the condition is not as similar to the adult version as was originally thought. One of the best examples of such confusion can be found in the area of reading difficulty where the adult onset (acquired) form was initially compared to the developmental (congenital) form. Although disagreement persists, most believe that adult dyslexia is not at all like developmental dyslexia. In the motor domain, we find similar problems arising from the word apraxia and its derivatives.

1.4.1 Apraxia, dyspraxia

The word "apraxia" is derived from the Greek word "praxis" meaning "to do" or "act". (see Table 1.3). In medicine the term "apraxia" was reportedly coined by Steinthal in 1871 and became a topic of the systematic studies by Liepmann at the beginning of the 20th century (Gonzalez Rothi & Heilman, 1997). The OED citation from Woodworth (Table 1.3) refers to damage of "the super-motor centers" (indicating the higher central nervous system) as the cause of apraxia. Within the adult literature, a whole range of different types of apraxia are described (e.g., constructional, dressing, ideational, ideomotor), all of which are attributable to localised cerebral damage. What these subtypes have in common, however, is that the performance of learned skilled movements is impaired, while muscle power and strength are unaffected (Oxbury & Oxbury, 1996). These "signs" can occur following a stroke, an invasive tumour or trauma such as a bullet wound in an adult or child. In contrast 'dyspraxia', 'developmental dyspraxia' and 'developmental apraxia' although evolving from the same root word 'apraxia' are interpreted rather differently when applied to developmental conditions.

Table 1.3 *Apraxia and Agnosia: OED (1991) Origin, Earliest Quotes and References*

Apraxia

From Greek Praxis meaning "to do"

Apraxia: Inability to perform purposeful movements; loss of ability to do.

1888 M.A. Starr in Medical Record XXXIV 497 title Apraxia and Aphasia; their varieties, and the methods...for their detection.

1922 R.S. Woodworth Psychology iii. 57 Injury to the "super-motor centers" causes loss of skilled movement, and produces the condition of 'apraxia', in which the subject, though knowing what he wants to do, and though still able to move his limbs, simply cannot get the combination for the skilled act he has in mind.

Agnosia

[Mod Latin from Greek meaning Ignorance]

Freud's term (zur auffassung der Aphasien, 1891) for loss of perception.

1900 Dorland Med. Dict. 27/2 Agnosia, loss of the perceptive power; loss of power to recognise persons or things seen.

One of the first writers to compare the adult and developmental forms of apraxia directly was Orton (1937). On page 47 he defined (adult) apraxia as: "loss of a previously acquired ability to carry out intricate skilled acts." On page 120 he then used the term in a heading: "Developmental Apraxia (abnormal clumsiness)" and continues in reference to children (p. 120) - "The fifth group of cases to be discussed is that of developmental apraxia (congenital apraxia). Such children are often somewhat delayed in learning even the simpler movements such as walking and running, and have great difficulty in learning to use their hands and to copy motions shown to them. They are slow in their attempts to button their clothes, tie their shoes, handle a spoon, and other simple tasks." Orton thus differentiates 'Acquired apraxia' from 'Developmental (Congenital) apraxia without really identifying the problem to be solved. With the prefix 'congenital' or 'developmental', apraxia was thereafter adopted into medical paediatrics.

So, what is the problem? It is important to note that Woodworth (Table 1.3), a psychologist, refers to apraxia as 'loss of skilled movement' and Orton similarly firmly defines acquired apraxia as a "loss of previously acquired ability." Orton then goes on to describe developmental apraxia as a "failure to learn new motor skills." This is the crux of the present controversy surrounding interpretation of terms derived from apraxia. Growing children, unlike children or adults who have suffered a neurological insult or pathology, have **not** lost ability but have not yet acquired skilled movement proficiency. Thus the question is what, if anything, does a child with a cerebral tumour, who has suffered neurological insult, which results in sudden inability to carry out specific actions voluntarily which they have already learned, have in common with a child who has more difficulty than his peers in the **development and acquisition** of movement competence? Whether one can justifiably extend an established neurological term, that is still in current use, associated with specific pathology, and apply it to developing children is arguable. However, the term was initially useful. A quick survey of any group of professionals familiar with children who have been brain injured, as a result of a car accident or tumour, for example, will reveal that no-one views these children as remotely similar to children who have **never** been proficient at movement skills.

Another problem that arises within this debate centres on the idea of dyspraxia as specifically a disorder in the **planning** of actions as opposed to the **execution** of skilled movement. This again has stemmed from adult neurology where constructional apraxia, for example, is manifest in difficulty planning how to assemble parts to make a whole where a spatial component is involved, in spite of having all the necessary motor control to make the necessary movements e.g., building a model, arranging a pattern, planning a drawing. In developing children, failure to build a model usually reflects **immature** function rather than **abnormal** function compromising planning. Words again provide confusion when 'executive function' within cognitive neuropsychology is found alongside 'execution of motor function'.

In yet another reference to adult pathology we find 'apraxia' linked to 'agnosia' (Collier & Adie, 1922). This link can also be found in the writings of Walton et al. (1962), and Gubbay et al. (1965) with Gubbay (1975, p. 41) referring to agnosia as an inability to recognise the significance of sensory stimuli, "actually perceptive

defects". In his influential volume on the topic, Gubbay (1975) used a string of terms which he defined strictly and his definition of a clumsy child remained the gold standard for over twenty years. On page 39, he writes "In the context of this report, the "**clumsy child**" is to be regarded as one who is mentally normal, without bodily deformity, and whose physical strength, sensation, and co-ordination are virtually normal by the standards of routine conventional neurological assessment, but whose ability to perform skilled, purposive movement is impaired". Gubbay goes on to say: "This type of **clumsiness** is designated by the neurological term **apraxia**. As praxis and gnosis are so closely allied and are interdependent in the performance of skilled movements a defect in one will result in disturbance of the other, either because of **impairment of integration** or **kinaesthetic feedback**. Both terms **'apraxia'** and **'agnosia'** can be applied to the manifest disabilities of these children. The term **'developmental'** implies a **congenital or early acquired defect or disorder** in the development of a particular function. The functions in this context refer to **gnosis** and **praxis** and hence the terminology **'developmental apraxia and agnosia'** can be justified as the precise nosology pertaining to the clumsiness of these children, or more succinctly **'developmental apraxic ataxia'**" (The present author's emphasis).

Gubbay's circumlocution embraces the causal concepts of impaired sensory integration favoured by Ayres and her followers (Ayres, 1972; Fisher, Murray, & Bundy, 1991) and impaired kinaesthetic feedback preferred by the Laszlo school (Laszlo & Bairstow, 1983). Gubbay illustrates the difficulty of deciding upon one term to describe the child who appears 'clumsy'. Linking words such as apraxia+agnosia+ataxia suggests a child who may present manifest difficulties in planning + perceptual knowledge + execution of smooth actions. We may be forgiven for pondering over the past thirty year's progress, or lack of, in understanding the sources of movement difficulties in these children.

Ayres (1972) in her classic book *Sensory Integration and Learning Disorders* devoted chapter 11 to 'Developmental Apraxia' which she described as a disorder of sensory integration which interfered with the ability to plan and execute skilled or non-habitual movements. Although it is unlikely that they ever met Ayres and Gubbay came to similar conclusions from quite different directions. Comparing their books (Ayres, 1972; Gubbay, 1975), there are many common sources in the bibliography. Gubbay refers to four publications by Ayres and also highlights the

work of Orton and Walton however Ayres does not mention Orton, Walton, or Gubbay, all of whom were European and not American. Geschwind (1975), cited by both the above authors also identifies apraxias as a group of conditions of disorders of **learned** movement. The term is also used by Roy (1978), Kelso & Tuller (1981), and Iloeje (1987).

The OED (1991) gives no entry for dyspraxia although the words dyslexia, dysphoria, dysplasia, dsypnoea, dysprosody and dystopia are listed. OED defines the prefix dys- as: "with notion of hard, bad, unlucky, etc. destroying the good sense of a word, or increasing its bad sense. In English, used in many words chiefly scientific, derived and compounded from Greek". The first use of the term dyslexia formed from German 'dyslexie' is given as 1883. 1886-8 W. R. Gowers in *Diseases of the Nervous System* is quoted as referring to "the cerebral symptom 'dyslexia'. A peculiar intermitting difficulty reading." Gower's reference to "the cerebral symptom 'dyslexia'" is interesting in the context of discussion of '**syndromes**' versus '**symptoms**' (see Chapter 2). The OED citation from the 1960 New Scientist, 15 Sept. 738/2 "Specific dyslexia"...deplorable term "word-blindness" should be avoided" (OED, 1991) provides a parallel with the discussions of the avoidance of derogatory labels linked with clumsy. Dyslexia, historically, shares with dyspraxia many similar problems in its recognition, interpretation, classification, the symptom/syndrome controversy and concepts of co-morbidity or co-occurrence (see Chapter 3).

'Dyspraxia', although well recognised as a medical term, was much later than the term 'dyslexia' in its entry into common English. There was certainly an entry in a 1965 Medical Dictionary (Dorland, 1965) "Dyspraxia: [Gr dyspraxia ill success] Partial loss of ability to perform coordinated movements" but only in 1997 does it appear in UK in The Oxford Dictionary of 'New Words' (1997).

"Dyspraxia. A disorder marked particularly by impairment of the ability to co-ordinate motor movements, and now associated with difficulties in reading and spelling....The term dyspraxia is recorded in medical literature from the early part of the last century, but it is in the 1990s that the term has made its way into the mainstream vocabulary. Dyspraxia is rapidly becoming as familiar as dyslexia in discussions of reading difficulties, although the degree to which its ready use reflects

an accurate medical diagnosis is still a matter for debate. Those suffering from dyspraxia are described as dyspraxic.... "Recently the newer syndrome of dyspraxia has replaced dyslexia in fashionable schools and educational circles as the explanation why little Freddie can't pass into Porridge Court" (Daily Telegraph 16 Nov 1995, p 31 cited by the Oxford dictionary of New Words, 1997)

In the UK, increasing public familiarity with the term 'dyspraxia' seems likely to have been prompted by the term's medical ring and similar sound to the already familiar word, 'dyslexia'. This was definitely the thinking behind the selection of the term by the group of parents who formed the Dyspraxia Trust in the 1980s (subsequently renamed the Dyspraxia Foundation). This group has stimulated the rapid rise in the use of the term in Britain although their information leaflet departs from the original meaning of the word and gives 'dyspraxia' a new and very broad definition:

"What is dyspraxia? It is an impairment or immaturity of the organisation of movement. Associated with this there may be problems of language, perception and thought. Other names. Clumsy child syndrome, Perceptuo-motor Dysfunction, Minimal Brain Dysfunction, Motor Learning Difficulty." (Dyspraxia Foundation, 1997).

Besides the Dyspraxia Foundation in UK there are currently many apraxia or dyspraxia support groups and parent networks with information accessible via the World Wide Web, e.g., Apraxia Kids (USA), The Dyspraxia Support Group of NZ, The Dyspraxia Association (S. Ireland). One of the major questions that emerges in this context is perfectly captured in the title chosen for a paper by Missiuna & Polatajko (1995) "Developmental dyspraxia by any other name: are they all just clumsy children?"

Two other terms worthy of passing mention with roots in adult neurology that have extended to the paediatric realm include 'Choreiform syndrome' and 'Gerstmann syndrome'. Several authors focused their research on the 'soft neurological signs' observed in children displaying 'clumsiness' (Schaffer, 1978). A mixture of positive signs such as retained reflexes and negative signs, for example absence of normal postural reactions were described. The child's movements mildly resembled the choreoathetoid and ataxic motor patterns found in cerebral palsy (Prechtl &

Stemmer, 1962; Wolfe & Hurwitz, 1966). Tests were devised to try to evaluate these motor patterns (Fog & Fog, 1963). Developmental Gertsmann Syndrome evolved from descriptions by Gerstmann (1940) of a syndrome of finger agnosia and lack of knowledge of right and left. This syndrome was later revisited and a suggestion made that clumsiness in children might be a form of 'Gerstmann syndrome' and it was prefixed 'developmental'. (Kinsbourne & Warrington, 1963; PeBenito, 1987). The term is rare in contemporary paediatric literature.

In summary, such commandeering of established medical terms, used and clearly defined within adult neurology in relation to acquired pathology, has caused a muddle when applied within the context of child development. The derivation from neurology has overemphasised 'clumsiness' from a narrow, medical perspective although it enabled a hitherto unrecognised problem to be placed 'on the map' and thus stimulated and attracted formal research. However, although many health professionals may be aware of the usage in neurology, educational professionals and lay people have never met this interpretation. Thus, in contrast to a lay term 'clumsy' which became a medical syndrome 'the clumsy child syndrome' we have a medical syndrome 'apraxia' being used as a lay term by parents.

1.5 Terms used to denote a continuum of brain damage - MBD (Minimal Brain Damage)

A quite different source of confusion, from that which attempts to relate adult neurology to developmental neurology can be found in the literature on infant development, the focus of neonatalologists, paediatricians and psychologists interested in the development of the brain both prenatally and after birth. This takes as its starting point the idea that there is no clear dividing line between a normal and an abnormal brain - particularly in the area of developmental disability.

> "From the standpoint of developmental diagnosis one might postulate that all children, whether first-born or not, actually suffer some degree of natal injury; but the vast majority of normal and fortunate infants have mechanisms of adaptation which result in prompt recovery. The minimal injury group consists of those children who make a slow or delayed recovery or who present persisting behavior residuals consequent upon inferred injury. Gesell & Armatruda." (1941, p. 232)

Schachar (1986, p. 19) cites Still (1902) and Tredgold (1908) as promulgators of the concept of brain damage as leading to disorders of behaviour generally and movement in particular. In the 1940s, Gesell and Armatruda, quoted above, provide one of the earliest examples of terms which imply causation rather than purely observational descriptors. For instance in the Introduction to their book on Developmental Diagnosis, they categorise injury into 'devastating', 'selective' and 'minimal' and most importantly note that "The child with only minimal injury needs the very same recognition and understanding, [as a child with selective injury who is obviously handicapped] and he too needs more than ordinary protection from stress and competition, particularly during the early years" (Gesell & Armatruda, 1941). Similar ideas are put forward by Knobloch and Pasamanick (1959) who refer to the notion of 'a continuum of reproductive casualty'. Perinatal insults can give rise to 'degrees of damage' from fatal cerebral impairment to lesser damage associated with later learning and behaviour problems. This led to the view that even when unequivocal neurological signs were absent disruption of the child's development might still occur. In the light of current research using state-of-the-art imaging techniques (Bos, Martin, Okken, & Prechtl, 1998; Jongmans, Henderson, de Vries, & Dub, 1993; Levene et al., 1992), Gesell and Armatruda's, and Knobloch and Pasamanick's comments seem particularly pertinent.

Gesell and Armatruda suggest that natal insult is the norm and that most children adapt and recover full function. However they do not expand on what they mean by 'full function' and whether this applies to physical and/or mental outcome. In contrast Strauss and Lehtinen (1947), clearly distinguish able children by their statement that they will pay particular attention to those cases of brain injury in children in which intelligence quotient remains at a normal level and suggest that dysfunction is related to specific perceptual processes. Lehtinen suggests that brain injured/damaged includes the clumsy group of children by her use of phrases familiar to past and future literature, such as awkwardness, lack of perceptual and motor integration, clumsiness, carelessness - "Much of the awkwardness of the brain-damaged child appears to be due to a lack of integration of perceptual and motor systems, as well as the failure of the visual perceptual processes to provide substantial and clearly structured patterns for the motor actions to follow. A primary function of vision is to direct movements. The clumsiness of the child in bumping into furniture, or banging a chair he is moving into doorways or other pieces of

furniture is not only because he is careless or awkward. Since his space world relationships are not accurately perceived they cannot serve as an accurate guide for his movements" (Strauss &Lehtinen 1947 p. 173).

Since the 1960s a deluge of terms flooded the field with words such as 'minimal', 'minor', 'mild' and 'cerebral', 'brain', 'neurological' and 'syndrome', 'dysfunction', 'delay' merged in various combinations: e.g., 'minor nervous dysfunction' (Touwen & Prechtl, 1970); 'cerebral minimal syndrome' (Frostig, 1971); 'minimal cerebral palsy' (Watter, & Bullock, 1987); 'minimal brain dysfunction' (Clements & Peters 1962; Abbie, 1974; Denckla, 1978; Kalverboer et al., 1978; Nichols & Chen, 1980); 'minor neurological dysfunction' (Touwen, 1979; Hadders-Algra et al., 1988); 'minimal neurological dysfunction' (Unwin, 1995); 'minor neurodevelopmental disorders' (Gillberg & Gillberg, 1989). Although the selection of terms to be combined often seems to have been almost random or simply due to personal preference, one can discern a systematic move away from the term 'brain **damage**' to 'brain **dysfunction**' e.g., Rutter (1977) uses the key phrase "brain damage syndromes" whereas five years later his choice of term is 'minimal brain dysfunction' (Rutter, 1982). These changes in emphasis reflect signs of a move toward appreciation of functional abilities rather than disease and impairment and are mirrored later in the development of the International Classification of Function (ICF; WHO, 2002).

Gubbay (1975, p. 35) cites Kramer and Pollnow (1932) as "amongst the original writers on the subject of minimal cerebral dysfunction". This term was commonly used in the 1960s (Bax & MacKeith, 1963) as an umbrella term for children with so-called 'soft' (i.e., non-specific) neurological signs plus functional difficulties in movement language, and attention. Paine et al. (1968) commented that conventional neurological examination usually showed no abnormality of the standard signs such as in the cranial nerves and reflexes, although a certain number of patients had extensor plantar responses or hyperreflexia. However, clumsiness was noted which was more likely to affect fine muscle coordination rather than gross functions of running, jumping and hopping. This heralds the concept of sub-types, which will be discussed later in the thesis.

The word '**dysfunction**' as oppose to '**damage**' or '**injury**' reflected consequences rather than causes of abnormality. Ingram differentiated a group of children without cerebral palsy who had what he termed 'chronic brain syndromes'. This included the child who lacked motor competence (Ingram, 1963). Illingworth (1963) also lists symptoms reported by parents of 'clumsy' children - "Teacher says he has difficulty with his pencil", "He turns his left foot in when he gets tired", "He can't pedal a bicycle". Illingworth continues "I regard these cases as examples of truly minimal cerebral palsy" and later adds "The importance of these cases is the fact that they would all pass at school age as normal children, and are therefore apt to get into trouble at school for clumsiness of movement, bad writing, breakages of glassware, and poor performance in physical training and dancing". Clumsiness here is seen as a continuum and no more than one end of the cerebral palsy spectrum, an issue revisited in the next chapter. MBD and its associated terms have mainly fallen from general use. A firm rejection of the term 'minimal brain damage' can be found in the proceedings of The 1962 Oxford International Study Group on Neurology. Participants included representatives from neurology, paediatrics, anatomy, ophthalmology, psychology, child psychiatry, physical medicine, public health, The Spastics Society, epidemiology, neuropsychiatry, neuropathology, neurochemistry, pathology, orthopaedics, and genetics: all voted for rejection of the term. Suggestions, made at the same meeting, for an alternative label to MBD were rather cumbersome: e.g., "minimal symptomology of brain damage", "minor brain disorders", "minor disorders of cerebral function", "Children with minor manifestations of cerebral dysfunctions". None of these labels was adopted as a suitable replacement for 'MBD' (Bax & Mackeith, 1963). A suggestion was put forward that there was need for more than one label; namely, one for precise medical use, a term for less precise administrative use and a colloquial language for general usage (e.g., with parents).

Firm rejection of the term 'MBD' can also be found in the current ICD10 (WHO, 1992) entry under Specific Developmental Disorder of Motor Function (SDDMF) (see Table 1.4) The entry includes the following comment: "The clumsy child syndrome has often been diagnosed as "minimal brain dysfunction", but this term is not recommended as it has so many different and contradictory meanings".

1.6 Geographical and professional individuality

In addition to the major sources of confusion discussed so far, two lesser sources can be identified. In the first instance, certain countries have consistently used their own terminology and seem resistant to change. Whereas geographical preference for a term may raise no difficulty locally, problems arise when a child moves outside that region to another country where the label used is not understood. The second source of confusion arises from the use of terms by one profession which are not understood by another. Surely, the bare minimum requirement is that a child's teacher and parent should know what his/her therapist is talking about.

Two examples must suffice to illustrate the problems raised by geographical isolationism. As noted earlier, one of the first clear references to children, who we might now describe as having DCD was made by Duprè and Merklen in 1909. They described a syndrome which they named "syndrome de déblité motrice" a term which remained exclusively French and was never adopted elsewhere. The idea that débilité motrice (motor debility) lay on a continuum between frank cerebral palsy and normal motor competence was not unique, however, and is a theme taken up in Chapter 2 of the thesis.

In the present day, Scandinavia stands out as being rather resistant to the influences of the rest of the world. There, the acronym DAMP is commonly used and understood as standing for 'deficits in attention, motor control and perception'. Proposed by the influential paediatrician Christopher Gillberg, the term has reportedly been in clinical use for about 20 years to signify concomitant attention deficit/hyperactivity disorder and developmental coordination disorder (Gillberg, 2003) and has the advantage of drawing attention to the fact that motor problems are often associated with deficits in perception and attention, a theme revisited later.

In parallel to geographical separation and national identity there is also professional separation and professional identity. Medical doctors, therapists, psychologists, educational professionals have well defined areas of expertise and are answerable to different professional bodies e.g., in the UK the General Medical Council (GMC), Chartered Society of Physiotherapists (CSP), College of Occupational Therapists (COT). Sometimes a term is linked to a professional group and is clearly understood by the members of the group. However as with communication across countries, use

of a term by one group may lead to lack of real understanding of the label outside a narrow circle. One example already given relates to 'dyspraxia', which from a pure medical neurology stance, is perceived rather differently to the lay interpretation by dyspraxia parent-support groups.

Another example of how one professional can be completely ignorant of what another means centres on the use and understanding of the term Sensory Integrative Dysfunction. Occupational therapists and to a lesser extent physiotherapists and speech & language therapists are familiar with the term 'Sensory Integrative Dysfunction'. This term was first introduced by Ayres who defines sensory integration in terms of the ability to organise and use sensory information (Ayres, 1972). She enlarges on this stressing that sensory integrative processes result in perception and other types of synthesis of sensory data that enable man to interact effectively with the environment. Ayres' adoption of the term sensory integration to indicate sensory information processing has become confused with her specific interpretation of sensory integrative **processes** and upon which the **techniques** of Sensory Integration **Therapy** are based. However for therapists unfamiliar with the main body of literature from psychological, physiological, educational and neurological sources, the model of integrating sensory information and motor output is often wrongly attributed uniquely to Ayres. With more widespread use of Ayre's sensory integrative therapy techniques internationally however, the term has become increasingly familiar to, although not always fully understood by, the general public. The term has not extended beyond the therapy profession and literature, and has received neither general acceptance of Ayres' concept nor elucidation of the SIPT symptom profiles outside this narrow field (Missiuna & Polatajko, 1995; Henderson & Barnett, 1998).

1.7 Present day

In parallel with the attempts to describe, classify and explain the many terms used to describe the children of concern here formal classification systems have developed with recognition at an international level. The purpose of such systems is to bring about some standardisation but this takes time and as Henderson & Henderson (2002) point out, the path toward uniformity and acceptance is a long and arduous one.

1.7.1 International Classification Systems

Two classifications systems exist, which attempt to provide precise definitions of childhood disorders as a standard yardstick for diagnosis: The International Classification of Diseases (ICD) published by the World Health Organisation (WHO) and The Diagnostic and Statistical Manual of Mental Disorders (DSM) produced by the American Psychiatric Association (APA). The history of these internationally accepted reference books and the relationship between the two is complex and yet again confusing. Publication of ICD preceded DSM but each manual currently has an entry relevant to 'clumsiness'.

The American Psychiatric Association's (APA) Diagnostic and Statistical Manual of Mental Disorders or DSM-IV states as one of its goals "to facilitate research and improve communication among clinicians and researchers" (1994, p. xv). The APA recognises that the manual is used by "psychiatrists, other physicians, psychologists, social workers, nurses, occupational and rehabilitation therapists, counsellors and other health and mental health professionals." The APA goes on to declare the number of advisers involved and their breadth of knowledge. It also alludes to the inclusion of consultations between the developers of DSM-IV and the developers of ICD-10 "for the purpose of increasing compatibility between the two systems" (ibid. p. xvi).

Throughout the history of medicine, there has been an undisputed desire for a classification of mental disorders. As noted in the introduction to DSM-IV, however, there has been less agreement on which disorders to include and the best method of organisation. The classification systems have varied according to whether they address clinical, research or statistical needs. An initial attempt on the subject recorded the frequency of one category "*idiocy/insanity*" from the 1841 census. By 1880 seven categories of mental illness were distinguished. Following World War II, and much influenced by the presentations of service men and veterans, ICD 6 (WHO, 1948) was published. This was developed in 1952 as DSM I by the APA. More explicit definitions/criteria as a means of promoting reliable clinical diagnoses, a multiaxial system and a descriptive approach devoid of aetiological bias, were incorporated into DSM II, III and III R (APA, 1975; 1980; 1987). Comprehensive literature reviews and systematic computer searches were used to try to achieve the goal of collating unbiased information from epidemiological, treatment and clinical studies to address nosology of mental disorders. However the difficulty regarding

definition and use of words is apologetically revealed under the heading "Definition of Mental Disorder" (p. xxi) "...The term mental disorder unfortunately implies distinction between 'mental' and 'physical' disorders that is an anachronism of mind/body dualism. A compelling literature documents that there is much 'physical' in 'mental' disorders and much 'mental' in 'physical' disorders. ...unfortunately the term persists in the title of DSM-IV because we have not found an appropriate substitute". "The concept of mental disorder, like many other concepts in medicine and science, lacks a consistent operational definition that covers all situations...different situations call for different definitions". These comments are pertinent to the current discussions regarding interpretation and operationalisation of entries for children with movement difficulties.

1.7.2 Comparison between Developmental Coordination Disorder (DCD) and Specific Developmental Disorder of Motor Function (SDDMF)

Both ICD-10 and DSM-IV provide separate entries for 'clumsiness' as a childhood disorder. Table 1.4 provides details of the entry and progression of the terms in each of these official manuals. The table acts as a reference for the next section of the present chapter, which examines similarities and differences between the entries for the two classifications. The reader is referred for further detail to several scholarly reviews (Henderson & Barnett 1998; Henderson & Henderson, 2002). The first obvious difference is in the choice of term 'SDDMF' and 'DCD'. Both refer to development and disorder but there has been argument whether 'motor function' and 'coordination' mean the same thing. One can be very pedantic about the use of a word and whatever the choice some argument and criticism will ensue. In context, both words relate to movement. However to the uninitiated the term 'developmental coordination disorder' does not necessarily presume muscle action. Whereas DSM provides a single term developmental coordination disorder, ICD-10 qualifies the SDDMF entry with several alternatives: clumsy child syndrome, developmental coordination disorder and developmental dyspraxia. While this may be helpful in drawing together a group of terms it also encourages continued use of disparate terms.

The ICD-10 and DSM-IV entries are broadly in agreement on core features of serious/marked impairment in the development of motor coordination, which compromises academic achievement or activities of daily living. ICD -10 indicates that the motor impairment is at least 2SD below age peers on a standardised test (fine

or gross motor). DSM gives no indication of a standardised measure to determine 'substantial' motor coordination impairment (Criterion A). The issues surrounding Criterion A will be raised in later chapters in the thesis. Although both manuals include a clause regarding cognitive ability there are differences in the suggested operationalisation. ICD-10 expects the administration of a standardised test and usually IQ below 70 is a criterion for exclusion. DSM is less specific and includes children with mental retardation but only if their motor difficulties are considered to be excessively impaired. From my clinical experience the following example might be useful. Most children with Down syndrome (Trisomy 21) present with motor delay and all have mental retardation. However two children with Down syndrome and comparable intellectual assessment may present with rather different motor skills due primarily to very different underlying muscle tone. Although children with severe mental retardation generally have poorer movement skills, some children with mental retardation are quick, agile, dextrous and show no lack of coordination.

ICD-10 and DSM-IV agree in their exclusion of a general medical condition. Here again, however there are significant differences. ICD-10 excludes any diagnosable neurological disorder directly related to visual or hearing defects. ICD comments on the acceptable presence of 'soft neurological signs' (choreiform movements, mirror movements and altered reflexes provided that these are symmetrical i.e., suggesting immaturity rather than localised pathology). DSM-IV specifically mentions excluded medical conditions such as cerebral palsy, hemiplegia, muscular dystrophy and pervasive developmental disorders (e.g., Autistic Spectrum Disorders). Page xxiii of the Introduction (APA, 1994) recommends that the specific diagnostic criteria in the manual "are meant to serve as guidelines, informed by clinical judgment' (sic) and are not meant to be used in a cookbook fashion". Diagnostic issues around interpretation of symptoms, syndromes and overlap of conditions are examined later.

Specific Developmental Disorder of Motor Function (SDDMF) has never become a commonly used term. Although Henderson & Barnett (1998) point out the combined term 'Developmental Coordination Disorder' had no precedent in existing literature, it has been adopted in line with DSM as a descriptive term, which is "as free as possible from particular theoretical commitments".

1.7.3 Towards Consensus

In addition to the power that the WHO and APA exert over clinicians generally, we can identify two other ways of working towards consensus on the terminology to be used in relation to the children of concern here. The first has been through organised meetings of experts, the second through the introduction of electronic coding of information, particularly in the health service.

One example of the expert panel approach took place in 1994 when a conference entitled "*Children & clumsiness: a disability in search of definition*" was held in London, Ontario Canada. Over 40 participants, from eight different countries spread across four continents, met following an invitation from The Department of Occupational Therapy, University of Western Ontario. At the conference the use of the word "*clumsy*" was absolutely rejected and it was decided that the best alternative term was 'Developmental Coordination Disorder' (DCD; American Psychiatric Association, 1987; 1994; Fox & Polatajko, 1994).

The second move toward consensus is associated with the emergence of electronic coding. Although both health and educational records are collected, only in the health domain do records have to take a standard form, which complies with national protocols. In the past, clinical coding of essential patient data was collected by health personnel, without the help of computers, and stored in filing cabinets that occupied increasing amounts of office space. Not surprisingly, the value of computers in other areas of information recording led to a move to develop an electronic patient record (DOH, 1999). One of the problems to be solved, of course, was the need for data from medical records to be translated from a mixture of colloquial, specialist, Latin and Greek terms into language that a computer can process to form an unambiguous electronic record.

Two parallel systems that were introduced in the 1980s are the UK-based Read System, and SNOMED founded in the USA (Health Service Journal, 1999). The Read codes were the brainchild of a Loughborough General Practitioner, Dr Read. A structured hierarchy of simple codes, primarily intended for use in general medical practice, grew to become Crown Copyright and widely implemented as Read Codes Version 2. They were adopted as far afield as New Zealand but not in the USA. In parallel with the British coding system, the College of American Pathologists

developed a rival system 'Systemized Nomenclature of Medicine' (SNOWMED-RT). SNOWMED-RT, employs a search engine that is able to formulate similar data for different codes. Quite recently, the Department of Health announced that the United Kingdom's National Health Service Information Authority (NHSIA) and the College of American Pathologists (CAP) had agreed to collaborate on a new system which would provide the essential building block for a common global computerised language (DOH, 1999).

The collaborative work on coding together with the DSM-IV and ICD-10 classifications comprise the formal language, which has important theoretical and practical implications, in the fields of both health and education. ICD together with International Classification of Function ICF (WHO, 2002) constitute the core classifications of the WHO Family of International Classifications (WHO-FIC). Whereas ICD and DSM classify disease and mortality the recent addition of ICF which focuses on health reflects a current move away from the medical model. There are on-going activities including web-based training in ICF and the production of internationally comparable disability tabulations which together with future planned publications of ICD-11 and DSM-V herald dynamic change in relation to classification and measurement of health and disease. A move toward uniform codes was shown by the publication of DSM-IV Text Revision (APA, 2000) which includes ICD codes bringing DCD and SDDMF under the same category - F82. In view of the London consensus and the recent streamlining of codes, DCD will be the term adopted henceforth in this thesis.

1.8 Conclusion

This chapter has reviewed the origins, history and development of terms relevant to the concept of a syndrome of 'clumsiness' in children. Although a move towards consensus is clearly indicated, it is also evident that this has not yet been achieved. This issue is taken up in an empirical study, which examined the terms, 'clumsy' 'dyspraxia' and 'developmental coordination disorder' as perceived by medical and non-medical professionals in the UK.

Table 1.4 *DSM and ICD Entries for DCD and SDDMF*

Developmental Coordination Disorder (DCD)	Specific Developmental Disorder of Motor Function (SDDMF)
DSM III R (1987) Developmental Disorders group includes discreet entry headed Developmental Disorder in Motor Skills: 'Developmental Coordination Disorder' (DCD) DSM-IV (1992) DCD listed under Axis I Clinical Disorders. Sub-headed: Disorders usually first diagnosed in infancy, childhood or adolescence. Sub group: 315.4 Motor Skills Disorder DSM-IV (2000) Text Revision Sub-group: F82 'Developmental Coordination Disorder'. (DCD) which is characterised by motor coordination that is substantially below that expected given a person's chronological age and measured intelligence". Main Features: "The essential feature of developmental Coordination Disorder is a marked impairment in the development of motor coordination (Criterion A). The diagnosis is made only if the impairment significantly interferes with academic achievement or activities of daily living (Criterion B). The diagnosis is made if the coordination difficulties are not due to a general medical condition (e.g., cerebral palsy, hemiplegia, or muscular dystrophy) and the criteria are not met for Pervasive Developmental Disorder (Criterion C). If Mental retardation is present, the motor difficulties are in excess of those usually associated with it (Criterion D). The manifestations of this disorder vary with age and development. For example younger children may display clumsiness and delays in achieving developmental motor milestones (e.g., walking, crawling, sitting, tying shoelaces, buttoning shirts, zipping pants). Older children may display difficulties with the motor aspects of assembling puzzles, building models, playing ball and printing or handwriting." Associated disorders may include Phonological Disorder, Expressive Language Disorder, and Mixed Receptive-Expressive Language Disorder. Prevalence is estimated to be as high as 6% for children in the age range 5 - 11 years. DCD is differentiated from motor impairments that are due to general medical conditions with specific neurological disorders where there is "definite neural damage and abnormal findings on neurological examination". It is stated that "if Mental retardation is present, DCD can be diagnosed only if the motor difficulties are in excess of those usually associated with the Mental Retardation". A diagnosis of DCD is not given if the criteria are met for Pervasive Developmental Disorder or Attention-Deficit/hyperactivity Disorder. Although these individuals "may fall, bump into things, or knock things over" this is "usually due to distractibility and impulsiveness, rather than to a motor impairment". There is no suggestion that this feature might have a motor planning or perceptual link or the notion of a common underlying cause.	ICD-9 (1978) Specific Delays in Development includes discreet entry 'Specific Motor Retardation' ICD 10 (1992) Disorders of Psychological Development groups together Specific developmental Disorders. (These disorders have in common (a) onset invariably during infancy or childhood; (b) impairment or delay in development of functions that are strongly related to biological maturation of the central nervous system; and (c) a steady course without remissions and relapses. In most cases, the functions affected include language, visuo-spatial skills, and motor coordination. Usually, the delay or impairment has been present from as early as it could be detected reliably and will diminish progressively as the child grows older, although milder deficits often remain in adult life). Sub group (F82) 'Specific Developmental Disorder of Motor Function' (SDDMF) Main features: "a serious impairment in the development of motor coordination that is not solely explicable in terms of general intellectual retardation or of any specific congenital or acquired neurological disorder (other than the one that may be implicit in the coordination abnormality). It is usual for the motor clumsiness to be associated with some degree of impaired performance on visuo-spatial cognitive tasks. The child's motor coordination, on fine or gross motor tasks, should be significantly below the level expected on the basis of his or her age and general intelligence. This is best assessed on the basis of an individually administered, standardised test of fine and gross motor coordination. The difficulties in co-ordination should have been present since early in development (i.e., they should not constitute an acquired deficit), and they should not be a direct result of any defects of vision or hearing or of any diagnosable neurological disorder. The extent to which the disorder mainly involves fine or gross motor coordination varies, and the particular pattern of motor disabilities varies with age. Developmental milestones may be delayed and there may be some associated speech difficulties (especially involving articulation). The young child may be awkward in general gait, being slow to learn to run, hop, and go up and down stairs. There is likely to be difficulty learning to tie shoe laces, to fasten and unfasten buttons, and to throw and catch balls. The child may be generally clumsy in fine and/or gross movements - tending to drop things, to stumble, to bump into obstacles, and to have poor handwriting. Drawing skills are usually poor, and children with this disorder are often poor at jigsaw puzzles, using constructional toys, building models, ball games, and drawing and understanding maps. In most cases a careful clinical examination shows marked neurodevelopmental immaturities such as choreiform movements of unsupported limbs, or mirror movements and other associated motor features, as well as signs of poor fine and gross motor coordination (generally described as "soft" neurological signs because of their normal occurrence in younger children and their lack of localising value). Tendon reflexes may be increased or decreased bilaterally but will not be asymmetrical". Although there is no diagnosable neurological disorder some cases have a history of perinatal complications e.g., prematurity or very low birth weight. "The clumsy child syndrome has often been diagnosed as "minimal brain dysfunction", but this term is not recommended as it has so many different and contradictory meanings". SDDMF includes: Clumsy child syndrome Developmental coordination disorder Developmental dyspraxia SDDMF excludes: Abnormalities of gait and mobility Lack of coordination secondary to either mental retardation or some specific diagnosable neurological disorder.

Chapter 2

Clumsiness as 'Symptom' or 'Syndrome'

2.0 Introduction

You clumsy thing! - at some point in time, most of us will have said this to someone nearby who has dropped something or bumped into us. As noted in the previous chapter, clumsy movement is quite normal in all of us, young or old. It is usually transient and often caused by an environmental factor which upsets the equilibrium of the perceptuo-motor system. In some children, however, the degree of clumsiness is so severe (and so persistent) that it crosses the boundary from normality into abnormality. In the previous chapter, the path toward recognition of a group of children with normal intelligence but who have exceptional difficulty achieving movement fluency, was traced historically. The chapter ended by welcoming the recognition of the syndrome by the WHO and APA but also warning of the constraints imposed by classification systems, which are being constantly revised as new information emerges. Some of the conceptual and practical problems still to be solved become the focus of this, and the next chapter.

In this chapter, the focus of attention continues to be on the **motor** problems experienced by a small proportion of children, whose IQ falls within the normal range. The primary problem addressed is whether there is enough evidence to support the idea that a distinct syndrome which stands alone and can be clearly and reliably differentiated from other childhood disorders really does exist. Following this discussion, the possibility that clusters of symptoms define distinct and meaningful subtypes **within** the syndrome is considered. Before attacking the fundamental question of whether DCD exists as a syndrome however, it might be useful to try to clarify some of the terms encountered in such discussions.

2.1 Definitions of symptom, sign, syndrome, disorder, disease, type, subtype

Table 2.1 provides examples of current definitions of the words, symptom, syndrome and related terms. A general dictionary defines a symptom as a "Change in body indicating its state of health or disease" and syndrome is defined as "Combination of several symptoms in disease". (Collins Shorter Dictionary and Thesaurus, 1995). In

medical communication, however, symptoms are subjective descriptors, which a patient tells a doctor, in contrast to signs which are objective evidence used to support a diagnosis. Medical jargon is filled with eponymous signs – Babinski sign (extension of the big toe), Trendelenburg's sign (dipping of the hip), Charcot's Triad (dysarthria, ataxia and tremor in multiple sclerosis). In turn, a set of signs **and/or** symptoms may form a syndrome. As always, however, a situation, which seems reasonably straightforward turns out to be less so and operationalising these terms poses difficulties, both practical and theoretical.

Table 2.1 *Symptom, Sign, Syndrome, Disorder, Disease: Definitions from a Medical and a General Dictionary*

Symptom: "A term applied to any evidence of disease. The term, physical sign, is generally applied to evidence of disease of which the patient does not complain but which is elicited upon examination" (Black's Medical Dictionary, 2002).
Symptom (Medicine) "A feature which indicates a condition of disease, in particular one apparent to the patient" (Concise OED, 2004).
Sign "A feature of disease as detected by the doctor during physical examination of the patient".
Syndrome: "A term applied to a group of symptoms occurring together regularly and thus constituting a disease to which some particular name is given: e.g., Cushing's syndrome comprising obesity, hypertension, purple striae and osteoporosis" (Black's Medical Dictionary, 2002).
Syndrome: "A group of symptoms which consistently occur together" (Concise OED, 2004)
Disorder: (medicine) "A disruption of normal physical or mental function" (Concise OED, 2004).
Disease: "Any abnormality of bodily structure or function, other than those arising directly from physical injury". (Black's Medical Dictionary, 2002).
Disease: "A disorder of structure or function in a human, animal, or plant, specially one that produces specific symptoms or that effects a specific part". (Concise OED, 2004)

For example, we might ask how objective or subjective signs and symptoms are and how exactly they relate to each other. A patient might complain of feeling weak and have difficulty hopping (symptom). This becomes a sign, once the weakness is

confirmed, by clinical examination and manual muscle testing e.g., using the MRC Scale (Medical Research Council, 1943). Conversely, a medical investigation may reveal deteriorating muscle strength on clinical signs and blood tests and only subsequently does the patient begin to notice and complain of symptoms of weak muscles. Physical signs may be quite easy to observe/elicit. They may be visible (a rash), audible (crepitations on respiration), palpable (a tumour) or detectable by smell (alcohol on the breath). In contrast psychological signs are often inferred indirectly from clinical impressions, interview or questionnaire (depression; measures of social interaction). Characteristics of symptoms and signs will depend upon the level of description and who does the describing. The patient may say 'I lose my balance when I turn quickly'. This may be described by others as a sign of basilar artery insufficiency (vascular or neuological opinion), in terms of poor co-contraction of the muscles providing stability (therapist or movement specialist) or dysfunction in the vestibular apparatus (audiology view).

As with the terms symptom, sign, syndrome, there may be disagreement on the nomenclature of a given disease and what differentiates a disease, a syndrome, and a disorder. There is often inconsistent use of terms and sometimes no very clear boundaries or rules for usage. For example, pneumonia is perceived as a disease not a syndrome or disorder because an inflammation of the lungs is clearly identified as the cause. In contrast, malabsorption syndrome, defined by a group of co-occurring signs including diarrhoea with steatorhoea and nutritional deficiency, has many distinct underlying causes e.g., pancreatic insufficiency, coeliac disease, cystic fibrosis, Crohn's disease. Another example, attention deficit and hyperactivity (ADHD) is referred to as a disorder and Asperger's as a syndrome and Tourette's seems to be interchangeable as syndrome or disorder.

It appears that a syndrome may be elevated to the category of a disease when the aetiology and pathology and natural history are clear. However many syndromes continue to retain the name of the person who initially identified the cluster of symptoms long after the disease process has been precisely documented; e.g., Hurler's Syndrome/disease (one of the group of lysosomal storage diseases due to a lack of α iduronidase, a mucopolysaccharide degrading enzyme) (Campbell et al., 2005, p. 48). A disease may also change its name thus what was previously known as disseminated sclerosis is now referred to as multiple sclerosis and juvenile chronic

arthritis (JCA) has recently become juvenile idiopathic arthritis (JIA). Confusion is compounded when a syndrome or disease is abbreviated: AS which may be Asperger's Syndrome or Ankylosing Spondylitis. TS which may indicate Tourette's Syndrome or Tuberous Sclerosis. DCD in the present thesis indicates Developmental Coordination Disorder but a literature search revealed that Donation after Cardiac Death is also abbreviated to DCD (Kaplan et al., 2004). Adding to the confusion, unrelated authors both named 'Bonnie Kaplan' published papers on DCD. One as referenced above on Donation after Cardiac Death and another on Developmental Coordination Disorder (Kaplan et al., 1998; Kaplan et al., 2004)!

It must also be remembered that the presence of one syndrome, disorder or disease in a person does not preclude the existence of another condition or conditions. A child with Down syndrome may also have unrelated asthma or have a related congenital heart defect or a medical condition such as leukaemia in later life. The idea of co-occurring, overlapping or associated conditions is pertinent to the present chapter but becomes a central theme in Chapter 3.

The ramifications do not end here as a disease or syndrome may also be classified into a number of sub-types and these may evolve and change over time. Type is defined as a class of things etc. that have common characteristics, and sub-type indicates a sub-division within the main class. Duchenne Muscular Dystrophy was originally identified as a single disease – Meryon's disease but later many different types were described and subsequently dystrophin gene mutations/deletions identified (Emery & Emery, 1995). This information lays the vital foundation upon which the search for prevention and treatment can be developed. Similarly it is necessary to identify the type of influenza organism in an epidemic in order to select an effective inoculation to protect the population. Rutter (1998), also underlines the importance, of not only clearly defining sub-types within developmental disorders but that putative sub-types must be validated by symptom profiles and ultimately, to evidence of positive response to specific intervention. Research in the field of reading difficulty has supported the existence of sub-types within dyslexia, and intervention aimed at improving reading may be specifically directed toward the underling processes, based on either auditory- or visual- based teaching methods. (Working Party, 1999).

2.2 Clumsiness as a symptom or syndrome- the clients' perspective

Clumsy movement and/or difficulty with the acquisition of age-appropriate movement skill is something which is noticeable to anyone who has close contact with a child. Parents, siblings, grandparents, teachers and peers may not make very accurate judgements about how delayed or different a child actually is but they will certainly be able to describe the behaviour in general terms. The children too are rarely unaware of their difficulties, especially once they are in school and able to compare themselves to their peers. Whatever, their relationship with the child, each will look at the 'clumsiness' from their personal perspective and reflect upon whether/how the movement difficulty is interfering with everyday function and hindering progress.

In most cases, concern usually starts at home when parents/carers notice that their child has more difficulty than siblings or peers with activities requiring coordination. Parents especially use the development of other children in a family as a means of identifying differences in rate of motor development. In some families, therefore, concerns about a child's clumsiness may start very early. The toddler who cannot use a spoon or fork, who seems to trip up on every step or kerb, walks into yet another cupboard door and is way behind his sisters and brothers, even those who are chronologically younger, usually makes the parents seek help. Such parents may also notice early feeding and chewing difficulty accompanied by speech which may be less distinct with hesitancy getting the words out. They may notice a slight floppiness in the infant and make comments such as his feet are really hard to push into his wellies. Many times they observe that rather than crawl their child bottom-shuffled and hand preference was not strongly established. At playgroup, they may notice the child seems afraid to attempt climbing up heights such as the slide or when he does get up a few rungs on the climbing frame has no idea how to get down again. A rather different stimulus for parents' seeking advice can be frequent falls resulting in grazes, torn, muddied clothes and sometimes repeated visits to Accident & Emergency.

In contrast, there are other families, with or without siblings for comparison, who either do not notice the tell-tale signs or simply consider them to be insignificant until the child enters formal education and a teacher comments on the difficulties and their effect on progress in school. Another reason why movement difficulties are not

considered crucial is that the child may have other difficulties, which seem more dominant at certain points in time. For example, delayed speech may take precedence over awkward walking. A child's tantrums may lead parents to seek help and advice but only later is the underlying cause revealed as frustration linked to the struggle to overcome clumsiness. Idiosyncratic behaviours may seem much more troubling to a family than clumsy movement. Parents may be so fully occupied coping with a child who perhaps freaks out whenever the vacuum cleaner turns on, or persistently lines up all his toy cars and hates anything being disordered, to notice that their child's fine motor skills are not developing normally. Parents may be overwhelmed at a child who they dub their 'little professor' who knows everything about space, or can recite the most unpronounceable dinosaurs or name every make of car. They may not be aware that the same child may be hopeless at catching a ball and have great difficulty playing sociably with peers.

Whenever a 'good' teacher encounters a child with movement difficulties, similar observations to those made by parents are made but from a somewhat different perspective (there are still a few teachers who consider clumsy behaviour to be the result of naughtiness). What teachers are concerned with is not only the lack of coordination per se, but how the difficulties affect progress in school in the context of a calm safe environment for learning and positive signs of potential for academic progress. In some cases, disruptive behaviour or lack of attention begins to compromise the smooth running of the classroom, a problem more common in boys than in girls. However, girls, less likely than boys to externalise behaviour may be overlooked. Teachers notice if letters and number are poorly formed and writing is untidy affecting legibility and presentation. They observe that the child takes a long time to complete work but sadly some teachers ignore the effort made by the child and may even punish a child by pressing for additional writing throughout break time. Teachers also see children moving around during PE and lack of skill makes the child stand out in the group. They notice when a child is falling in the playground or challenging safety but may perceive this as lack of control and fool hardiness. Although friendly social behaviour is apparent in the playground children who are loners or who prefer to chat to the teacher rather than join in physical play with peers may rouse suspicion only in experienced professionals.

From the child's perspective, the contrast with brothers, sisters and class-mates is often painfully obvious. Whereas they seem to manage to learn new skills without difficulty everything for him is a struggle and although he catches on one day on another occasion success evades him. When children discuss their problems with me in clinic they very often mention that they stumble and fall over and feel uneasy in the school playground. They say that their handwriting is messy or too slow and that it is hard to get stories onto paper. They say all their friends are better at sport and that they wished that they could catch a ball. They bemoan the fact that they are last to be chosen as a partner in P.E and never get picked to be in the team. Another aspect that bothers the child is inability to tie shoe laces or do up zips and buttons which often results in being ridiculed. They say they feel stupid when they cannot ride a bicycle, or ask for help in learning to skip or to do hop-scotch. Some children are able to verbalise their feeling of anxiety or anger when told by parent or teachers that they are lazy, careless or not trying. The child may use a strategy of avoiding physical activity but soon loses self-confidence and may make remarks such as "I'm hopeless at everything" "I'm a failure". The ever present yardstick for their symptoms is their friends, siblings and peers.

The many and varied symptoms reported above are not sufficient to differentiate between clumsiness that is signalling DCD and lack of movement competence which has another, quite different cause. The job of the professional is to tease out the possible explanation(s) for a child's difficulties, by interpreting reported symptoms and objective signs. This can then lead to differential diagnosis and appropriate intervention. Ideally a multi-professional team will work together in this process with each contributing their own particular expertise. Even when well-trained professionals are involved, however, the route to a diagnosis of DCD may be a long and tortuous one involving investigation at perhaps one or more specialist clinics in addition to therapy and educational assessment. Whether a medical cause is identified or a diagnosis made, may depend upon the depth of investigation, the sophistication of diagnostic technology, the number and severity of signs, symptoms and the degree to which function is disrupted within the child's environment. There is a medical aphorism that 'a normal child is one who is under-investigated' or as Rosenbaum (2006) recently reiterated "It has been said that 'normal' refers to people who haven't had enough tests!" Locality and resources will also determine both over- and under- investigation (Dunford & Richards, 2003). Eventually, however

there are some children who receive a diagnosis of DCD (or a local variant still in use) who present with a whole range of difficulties which are both motor and non-motor. In what follows the focus is primarily on the motor components while recognising the existence of other features (see Chapter 3).

2.3 Clumsiness as a 'symptom' or 'syndrome' – diagnostic issues

The issues surrounding the diagnosis and characterisation of DCD are discussed under five headings. These headings have been chosen to reflect not only the practical problems still to be solved but also to illustrate the theoretical issues that remain outstanding in this field.

- Clumsiness as the lower end of the normal continuum.
- Clumsiness as a 'symptom' of a known medical condition.
- Clumsiness as a mild form of cerebral palsy.
- Clumsiness as a unitary syndrome
- Clumsiness as a condition with clearly definable sub-types.

2.3.1 'Clumsiness' as the lower end of the normal distribution

DCD has often been referred to as a hidden handicap because there is no disfigurement or paralysis and the child looks normal. A few people perceive these children as simply falling at the lower end of the normal continuum and by implication suggest that there is no such thing as DCD. However this perception is often voiced by those who compare the child with more overtly multiply handicapped children and does not stand up to close scrutiny. Where might the idea come from and what are the issues?

There is no doubt that typically developing children mature at different rates, along slightly different trajectories, which are all within the normal limits. Some children walk at seven months, others not until over two years. Some children walk early but speak late. There are undoubtedly suspect children who appear to show early problems and then develop perfectly normally. The causes of this variation may be genetic, environmental or an interplay between nature and nurture so that it is important to consider many aspects of normal variability when deciding whether children fall out-with the norm at different ages.

For example, normal variation occurs in body type and build and this alters during development from infancy to adulthood with periods of particularly rapid change. Anthropomorphic variability relates to body mass index (height weight ratio), fat deposits, and musculo-skeletal flexibility. A high body mass index (BMI) may influence performance on explosive muscle action (jumping) and ability to maintain centre of gravity over base of support as in standing balance. If a child has an overly flexible musculo-skeletal framework (increased muscle and joint laxity or hypermobility) there are increased ranges of movement, which affect both postural control and manipulation tasks. These factors may mimic or compound the effects of DCD but are not in themselves the core aetiology.

Children vary in the opportunity to practise their movement activity. Driven either by parenting culture or the child's inherent make up an infant may lack experience because he/she is carried, pushed in a buggy, spoon fed and buttoned into clothes, thus allowing him/her to avoid developing a repertoire of adaptive movement skills. Increasingly, too, children from different cultures enter the UK education system and arrive with rather different 'norms'. A child from Holland, where bicycle-riding is expected to be accomplished early, will be more likely to be a proficient cyclist compared to UK contemporaries. A child arriving from Somalia usually has little or no experience of throwing and catching a ball. A child from China will handle chopsticks with a dexterity that leaves even UK adults amazed. Thus normality is partly defined in the context of a population which is culture specific.

Annell (1949), mentioned in the previous chapter, made the first explicit reference to clumsiness, falling at the lower end of normal development. She suggested that motor infantilism was due merely to a lag in motor development but not essentially abnormal motor development per se. Others have also claimed that at least some of the group described as clumsy reflect normal variability and are considered to fall at the lower extreme of the normal distribution. This is the view that has led to so many parents being told "Don't worry s/he will grow out of it" (Hall, 1988). However, there are now many studies which show that most of these children do not grow out of their difficulties at all (e.g., Gubbay, 1975; Losse et al., 1991; Cantell et al., 1994; Brown, 1996). One of the most recent, a retrospective study of adults with a diagnosis of DCD or history suggestive of childhood DCD, provides ample evidence

of the fact that motor difficulties can continue across the lifespan and have quite devastating effects (Cousins & Smyth, 2003).

Although the balance of opinion is that the majority of "clumsy" children do not simply fall into the lower end of the normal continuum, there are clearly times when it is difficult to be certain. This is especially true at younger ages. It is important, therefore, that an infant showing possible early signs of DCD should be carefully assessed and their progress monitored. Equally important, however, is awareness of the consequences of rushing to apply a diagnostic label such as DCD at the pre-school stage or even at school entry. Just as the previous section highlighted differences in children from different cultures there is marked variation in children's prior experience as they face 'day one' in reception class. Some arrive in school unable to cope and stand out in many ways. Some children have always had a parent beside them and never faced the world independently. The stress of several hours in a school classroom with 'strangers' is not insignificant. Some children are barely out of nappies, may still be drinking from a bottle and cannot cope especially alongside children at the other extreme who arrive confident, independent and already tackling writing and reading. But the children who appear infant-like and unprepared for school usually 'recover' relatively quickly once they settle and adapt to the school environment with support from understanding teachers. These children are not the subjects of the present thesis and the idea that children with true DCD simply lack experience or fall at the low end of the normal distribution can be dispensed with.

2.3.2 Clumsiness as a 'symptom' of a known medical condition

One of the major problems which faces professionals in the identification of DCD is to eliminate other **known** medical conditions in which clumsiness of movement is a common feature. During assessment the experienced physiotherapist remains constantly aware of the many alternative diagnoses, some very common and others rare, that must be considered. During the initial clinical assessment of such children the physiotherapist uses a combination of formal assessment tools, clinical experience and judgement to interpret and differentiate between a range of signs and symptoms in order to help in the diagnostic process. Referral details will provide important information and when the child has already undergone medical investigation a diagnosis may be established. Frequently, however, the child may arrive with sparse

medical details - just a brief request to assess clumsiness or frequent falls or in the case of direct referral the physiotherapist will be the first port of call.

The present researcher has had a unique opportunity through work at two special centres: GOSH, a world renowned specialist paediatric hospital, and SENSE (formerly the National Association for Deaf Blind and Rubella Damaged), a charity devoted to dual sensory impairment. This combined with community work in clinics and all types of schools provided over a 30-year period from 1975-2005, the opportunity to observe and assess several hundred children. All of these children were referred for physiotherapy assessment of symptoms of clumsiness. Table 2.2 is not an exhaustive list but serves to illustrate the many different conditions that may feature significant lack of motor coordination. For convenience the list is presented alphabetically.

Table 2.2 *Conditions that may Present with Symptoms of 'Clumsiness' in Children*

Asperger's Syndrome	Hypothyroidism
Attention Deficit/ Hyperactivity Disorder	Impaired hearing/vestibular dysfunction
(Benign) Joint Hypermobility Syndrome	Juvenile Idiopathic Arthritis
Cerebral Leucodystrophy	Metabolic diseases
Cerebral Palsy	Metatarsus varus
Child abuse	Muscular dystrophy
Chronic Fatigue	Neurofibromatosis
Conduct Disorder	Oppositional Defiant Disorder
Congenital hypotonia	Osgood Slatter
Congenital Rubella	Perthes Disease
Conversion Syndrome	Pes Planus
Cytomegalo Virus; Toxoplasmosis	Post Meningitis; Post Encephalitis
Depression	Posterior fossa tumour
Down Syndrome	Schizophrenia
Dyslexia, dyscalculia, dysgraphia	Sex Chromosome abnormality e.g. XXY
Dysregulation syndrome, dysautonomia	Side effect of medication
Ehlers Danlos Syndrome	Specific language impairment
Epilepsy	Spinal dysraphism
Fibromyalgia	Spinal Muscular Atrophy
Foetal alcohol spectrum disorder (FASD)	Tibial torsion
Fragile X Syndrome	Tourette's Syndrome
Global delay – mental handicap	Tuberous Sclerosis
Haemophilia	Vestibular hypoplasia
Heredity Sensory Motor Neuropathy	Visual Impairment
Hip anteversion; hip dysplasias	William's Syndrome

In what follows, some of these conditions are now examined further to illustrate how the process of differential diagnosis takes place. For convenience the conditions are loosely grouped under the headings of the systems that might be affected, with a broad distinction being drawn between physical and psychological conditions. In the present chapter, the focus is on diagnoses which have a distinct physical dimension, primarily affecting the musculoskeletal and neuromuscular systems directly. In the next chapter the interest turns toward psychological and psychiatric conditions.

Musculoskeletal system:- orthopaedics and rheumatology clinics: Parents/carers often become worried by their child's bow-legged or pigeon-toed gait. Sometimes they are also concerned that their child is unfit, weak (slouching over work, mouth breathing, running with a heavy elastic gait) and clumsy. On other occasions the child complains of pain in the legs, perhaps at night preventing sound sleep or after exercise. In some cases, initial referral for these symptoms may be to an orthopaedic or rheumatology consultant but in many instances, parent/carers detail the symptoms during the physiotherapy assessment of general clumsiness.

What do these signs/symptoms mean? A range of mild movement symptoms is related to alignment of bones and joints. Physiotherapy examination may show exaggerated rotation of lower legs (tibial torsion), flat feet (pes planus) and knock knees or bow legs (genu valgum/varus) that may appear to relate to falls. However these symptoms are usually transient features of immature gait (the normal sequence of hip alignment as upright stance is established) and usually all that is required is reassurance. In contrast, pain and inflammation of joints often signifies arthritis or bleeds into joints in haemophilia. The physiotherapist will look at the range of movement and for any signs of inflammation around joints or pain on moving joints, especially if localised or occurring in the early morning. These signs would make DCD less likely. While these conditions may give rise to clumsy symptoms, such children need different treatment.

Delayed motor development and clumsiness is frequently associated with ligamentous laxity and joint hypermobility, which compromises postural control. Signs include slight reduction in muscle tone (hypotonia) in the core postural muscles of the trunk and girdles. Laxity in the body framework may be due to one of the heritable disorders of connective tissue (Marfan Syndrome, Ehlers-Danlos

Syndrome, Osteogenesis imperfecta and Benign Joint Hypermobility Syndrome/BJHS). Once again, these conditions require a different approach to that taken with children with DCD. However, a number of recent reports have highlighted the frequency of flexibility in children with DCD (Kirby et al., 2005) and 'clumsiness' in children attending a hypermobility rheumatology clinic (Adib et al., 2005), suggesting a degree of overlap that requires further investigation (see Study 5).

Neurological system: neurodevelopmental, neuromuscular or neurosurgical clinics: When a GP notices that a child is abnormally weak a referral will be made urgently to a specialist clinic, so that very serious and life threatening conditions such as cerebral tumours, muscular dystrophy, may be investigated and confirmed or excluded. However, the early stages of several neurological or muscular conditions may be less overt and present with apparent benign clumsiness such as tripping up, difficulty with fine or gross movement, awkward ungainly actions, which may lead a child to see a physiotherapist. The physiotherapy assessment focuses especially on muscle tone, power and movement patterns. Markedly abnormal muscle tone or movement patterns, asymmetrical mirror reactions or reduced muscle power and fatigue especially if reportedly worsening should set alarm bells ringing. Clumsiness may be the initial symptom of several potentially deteriorating conditions such as a cerebral tumour, neuropathy, muscular dystrophy or neurofibromatosis (NF1). A child, especially a boy in school, who cannot even jump with both feet should be investigated for a possible muscle problem such as muscular dystrophy. Weakness of small muscles of hands or feet when other muscles appear normal is likely to indicate something other than DCD. Similarly, birthmarks and freckling may be indicative of NFI or unusual dimples indicative of spina bifida occulta.

Sensory systems: ophthalmology and audiology clinics: Parents and teachers sometimes comment on a child having difficulty listening to and processing instructions (e.g., in PE). Further enquiry then reveals that the child has had vision and hearing tests and that there is a history of glue ear, grommits, operation for strabismus etc. The 8th cranial nerve not only functions as the organ of hearing but also of vestibular function via balance receptors within the inner ear (utricle, saccule and semicircular canals). These are essential through stimulation of antigravity muscles for maintaining the head (with its sense organs) exactly oriented upon a stable body. Thus any dysfunction can compromise balance, muscle tone and upright

posture. During assessment the physiotherapist notes how the child listens, communicates and uses his/her vision. Clumsiness may be symptomatic of visual impairment disrupting precise movement judgement. The physiotherapist, will test body postures in a way that may reveal great difficulty in positioning limbs accurately unless the limb is directly within the visual field. Some children are particularly alert visually (parents say that the child notices everything) yet the child appears less tuned in to sensing how his body moves. How exactly these conditions fit into the DCD/not DCD picture has not been determined.

Homoregulation: metabolic; endocrine; genetic; immunology clinics. Parents/carers frequently comment that their child is disorganised, overactive, does not seem to listen and is easily distracted in addition to being clumsy and perhaps having difficulty with school work. When the presentation is complex affecting function in several domains the background history may also be quite complex. Medical records provide a variable amount of information and parents/carers are usually able to fill in missing details. Clumsiness may be symptomatic of residual effects of common infections, such as rubella, cytomegalo virus, toxoplasmosis, encephalopathies and meningitis. These can have devastating effects: the child may be severely hearing and/or visually impaired, paralysed or all of these but many mild or sub-clinical infections pass unrecognised - recovery is luckily complete or is it? Residual effects of such insults may leave no more trace than mild movement incoordination or clumsiness.

Genetic abnormalities or effects of environmentally derived substances may give rise to movement difficulty. It is a symptom observed in many chromosome abnormalities such as Down, Williams, Fragile X and Turner syndromes. In fact whenever muscle tone is altered motor function is likely to be affected. Substance misuse (alcohol, drugs) are known to affect motor control in the infant and clumsiness is one feature of foetal alcohol spectrum disorder (FASD). The child's features are observed for dysmorphic signs such as unusually low-set ears, hand or foot anomalies which may be telling signs. Other toxic substances, besides alcohol and drugs, such as lead may also be associated with clumsiness.

Finally, endocrine function is essential to maintain skeletal muscle metabolism, and hormone deficiency e.g., congenital hypothyroidism due to a deficiency in thyroid

hormone produces a myopathy. Many nutritional deficiencies (e.g, vitamin D and protein-calorie malnutrition) also cause muscle weakness. Additionally, certain medications e.g., for seizures, asthma, may produce side effects symptomatic of clumsiness. Thus, it is always important to check whether a child is on medication and especially whether medication (e.g., Methylphenidate) has been taken prior to assessment as this may affect the reliability and validity of a standardised test.

In summary the above section has focussed on the procedures adopted by the **physiotherapist** (and other colleagues) during clinical assessment of children described as 'clumsy'. Although many more examples could have been given, It is hoped that the above are sufficient to illustrate that the process is not a simple one. 'Clumsiness' is a symptom of many different medical conditions, some of which are not easily distinguished from DCD. Moreover, it must be borne in mind that co-occurring conditions are not unusual and a child may have, for example, tibial torsion and unrelated DCD which features poor manual dexterity.

2.3.3 Clumsiness as a mild form of cerebral palsy

In complete contrast to the idea that 'clumsy' children simply represent the low end of the normal continuum, the idea that they suffer from a mild form of a sometimes extremely debilitating condition, cerebral palsy, has also been expressed many times. As noted in chapter one, the idea was suggested nearly a century ago by Duprè and Merklen (1909) of Paris who noted that just as mental debility ranged in severity from severe to mild with idiocy representing the most profound form, motor debility lay on a similar continuum of intensity, with severe spasticity or athetosis at its extreme end. By implication, therefore, Dupré and Merklen suggested that a mild or form fruste, of motor agenesis, could be identified. They labelled this a syndrome of motor debility – 'syndrome de débilité motrice'.

> M. Dupré n'a eu en vue, dans la description du syndrome de la débilité motrice; que les formes supérieures, pour ainsi dire frustes, de l'agénésie motrice; mais la variété la plus complète de cette agénésie est réalisée par le syndrome de Little qui, véritable idiotie motrice, est aux formes incomplètes et frustes de la débilité motrice, ce que l'idiotie est à la débilité mentale. (Duprè & Merklen, 1909, p. 1074)

Jumping forward to the 1960s, we find a number of frequently quoted papers which propose that 'clumsiness' can be conceptualised as a mild form of CP, with the

publication by Illingworth (1963) being one of the most notable. At a conference on child neurology, Illingworth (1963) described symptoms and signs in a series of 27 cases selected from a cohort of 500 children with cerebral palsy seen by him at Sheffield Childrens' Hospital under the title 'The Clumsy Child' (Table 2.3)

Table 2.3 *Symptoms and Signs Reported by Illingworth (1963, p. 27)*

Symptoms (Reported by mother)	Signs (Elicited by the paediatrician)
Falls a lot; cannot run; bruises on legs	Abnormal unsteadiness in standing on one leg (all cases)
Slow at doing anything with hands Clumsy	Slight hypertonia with minimal signs of involvement of the pyramidal tract (8 cases; 2 cases also showed ataxia)
"Writes ever so queer" Has difficulty with his pencil; bad writing	Very slight ataxia or tremor on building a tower of cubes (all cases)
"Can't keep up with the others, he lags behind, so that they won't play with him" Cannot pedal a cycle.	Slowness and abnormal pattern of movement in standardised tests of repetitive movements involving the use of fingers (e.g., transfer of beads, threading, buttoning clothes)
"Turns his foot in when he gets tired" Poor performance in PT/dancing	Minimal athetosis (one case)

As the table shows, the symptoms reported by parents are identical to those now associated with children with DCD. According to Illingworth, what distinguished these children from those with 'overt CP' was 1) normal gait; 2) minimal neurological signs on careful testing; and 3) no history of possible causative postnatal disease such as encephalitis. He then goes on to point out that different children showed minimal signs of different forms of cerebral palsy, such as spasticity, athetosis and ataxia. Critical to this discussion, of course, is how one defines 'minimal'. However, a discussion of what is meant by 'soft neurological signs' would require a chapter in itself. Suffice it to say, that the measurement and interpretation of such signs is extremely controversial. Many of the children who turn up in a physiotherapy clinic show these signs but what they mean is less and less clear. In his summary of these cases, Illingworth describes the children as examples of '*truly minimal cerebral palsy*' and suggests they fall into the group described by Gesell & Armatruda (1941) as caused by '*Minimal Birth Injury*'.

Whereas Illingworth took the view that all children described as clumsy were simply cases of mild CP, others have suggested that this is only true of **some** children who

show these symptoms. For example, Henderson and Hall (1982) studied 16 children (aged five to eight years), from four mainstream schools who were identified as 'clumsy' by teachers. Very careful blind testing of soft signs differentiated the 'clumsy' children from age and gender matched normal controls. Although none of the children demonstrated overt CP some may have fallen on the fringe of such a diagnosis since 94% of the 'clumsy' children had a history of significant events in their medical history. The study further suggested three groups of children, those with an isolated motor impairment, a group where poor motor competence was combined with concern about academic progress and behaviour and a third 'intermediate' group that was less easily classified. More recent longitudinal brain imaging of very low birth weight and pre-term babies (Jongmans et al., 1998) and full term infants (Barnett et al., 2002), support the idea that at least a sub-group of children who might bear the label DCD fall within the fuzzy grey borderline of CP. This observation is reflected by Hadders-Algra (Hadders-Algra et al., 1988; Hadders-Algra & Gramsbergen, 2003) who suggests 'simple' and 'complex' dysfunctional groups (resembling CP) in her follow up studies of the Groningen Perinatal Project.

When considering the question of DCD versus mild CP as a label for the children at the centre of this thesis it is informative to turn briefly to another literature, that on cerebral palsy itself. For many years, there has been debate about the usefulness of this term and it is now generally conceded CP is not one, single condition and should therefore be referred to as the cerebral palsies. In a seminal paper on the classification of CP, Alberman and Stanley (1984) emphasise the many pitfalls that are presented to epidemiologists in the study of cerebral palsies. Several points that they emphasise could be applied equally well to the present discussion of DCD. For example, (i) they repeat the fact that cerebral palsy is not a single condition but a miscellany of clinical syndromes with various causes, (ii) they note the fact that the presenting picture is complicated by the interwoven effects of reproductive hazards with social and biological factors, and (iii) they present data on the difficulties encountered in the classification of the individual syndromes within cerebral palsy. Cerebral palsy has been grouped into at least five main sub-types (Cans, 2000; Phelps, 1949). In addition impairment is classified topographically. Thus hemiparesis predominently affects one arm and leg and diplegia affects primarily the legs. Finally all categories may be placed on a continuum of severity of impairment from severe to mild. Whether one uses a topographic approach or otherwise one may hypothesise

that some children with DCD might belong to the mildest end of the continuum and show features or traits of any of the CP subtypes. For instance, a child who demonstrated mild subtle signs of inflexibility in the lower limbs may suggest minimal or residual symptoms of diplegia. (See Case 4, Chapter 8). Once again, however, we are faced with the difficulty of defining concepts like 'mild' evolving reliable measures with well validated cut-off points.

This section has considered the proposition that 'clumsy' children lie at the mild end of a continuum within cerebral palsy. In its pure form, this hypothesis was rejected and a modified form accepted. This states that **some** children with movement difficulties may indeed be well described as 'mild CP' but certainly not all. Put the other way round, one might hypothesise that DCD as a syndrome encompasses a sub-type with CP traits but distinct from CP. How this particular approach moves forward, however, is still uncertain. Perhaps the current rapid advances in brain imaging techniques will help. As Pellegrino (1995) suggests, however, the present era is one of *"descriptive diagnoses and etiological agnosticism"* and the new paradigm for cerebral palsy must recognise that *"cerebral palsy is an eclectic diagnosis which is descriptive of disability rather than impairment"*. Where does that leave the discussion of DCD?

2.3.4 Clumsiness as a unitary syndrome

Syndrome: "A term applied to a group of symptoms occurring together regularly and thus constituting a disease to which some particular name is given: e.g., Cushing's syndrome comprising obesity, hypertension, purple striae and osteoporosis" (Black's Medical Dictionary, 2002).

Syndrome: "A group of symptoms which consistently occur together" (Concise OED, 2004)

So far, the focus in this section has been on the process of **exclusion** rather than inclusion - what DCD is **not** rather than what it **is**. There were two reasons for proceeding in this way. The first was simply to emphasise the fact that "clumsiness" of movement is not a rare phenomenon. Even when one excludes the clumsiness that is associated with normal development, one finds that lack of coordination is a **symptom** of a whole range of medical conditions. The second, and more important reason was to show that distinguishing DCD, as **a syndrome**, from these other

conditions is not a straightforward matter. Indeed, accepting from the start that the boundaries are blurred, will not disappear and somehow have to be dealt with in any classification system, seems like a good starting point.

In the UK, John Walton has the dubious reputation of creating the "Clumsy Child Syndrome" (Walton, 1961; Walton et al., 1962). This unfortunate term was first coined in the 1960s when Walton led the field in publishing the first of several papers relating to this syndrome which were then summarised in a BMJ editorial (Editorial, 1962) which referred to the work of Walton (1962), Prechtl and Stemmer (1962), and Illingworth (1963). These publications were pivotal in the direction taken in conceptualising the syndrome of childhood clumsiness in the UK, and are of great interest when one considers the syndrome, DCD, as it is now described in formal classification manuals, DSM and ICD (APA 1987; 1994, 2000; WHO 1992).

In DSM-IV, DCD is listed among "Disorders usually first diagnosed in infancy, childhood, or adolescence" and defined as a "Motor skills disorder". Four diagnostic criteria are then laid out. Each of the four criteria, however, presents some problems of interpretation (Henderson & Barnett, 1998; Rispens et al., 1998). In theory, Criterion A (marked impairment in motor coordination) should be relatively easy to implement. A child may be compared against norms using reliable and valid objective methods. However, what the literature reveals is that agreement between the various standardised tests is not strong and that although each may be reliable in its own right each may identify a slightly different group of children. Criterion B (significant interference with academic achievement or activities of daily living (ADL) is somewhat more problematic as the perception of significant may be different in the eyes of the child, parent, teacher or examiner. In practice, however, broad agreement on whether or not the child's difficulties are seriously compromising developmental progress can generally be reached. In contrast, this is not necessarily so when it comes to Criterion C (exclusion of medical conditions), which critics cite as being the most difficult to operationalise. As noted above, a substantial number of medical conditions exist which are not easily excluded without expert knowledge and/or objective tests. Finally Criterion D, specificity of motor impairment in relation to IQ, is beset with problems. Not only is it difficult to operationalise because only certain professionals are qualified to use IQ tests and others must rely on school reports of the child's intellectual ability, but more

fundamentally the whole concept of the specificity notion has been called into question (Rispens et al., 1998). On top of all of this, there are also problems when a child presents a complex picture which seems either to be on the fringe or cusp of several diagnoses or, conversely, does not clearly fall into any one DSM code. In the light of these difficulties one might ask (a) why should anyone continue to believe that DCD exists, and (b) whether the DSM criteria are worth refining. A historical perspective may help.

The BMJ review, entitled "Clumsy Children" (Editorial, 1962), cites studies from four centres, which reported on different groups of clumsy children. These included Annell (1949), Walton et al. (1962), Prechtl and Stemmer (1962), and Illingworth (1963). The last presented his paper "The Clumsy Child" at the 1962 Oxford Study Group on Minimal Cerebral Dysfunction (p. 26/7) alongside Walton's "Clumsy Children" (pp. 24-5) and a paper entitled "Spontaneously arrested hydrocephalus" (p. 28) by Hagberg (1962) who described a slightly different group of children 'many of them known as clumsy'. In addition, other aspects of clumsiness were emphasised by Paine (1962). Of these, there is space here to discuss just two.

Beginning with the often-cited Walton study (Walton et al., 1962) it is surprising to find that this paper contained descriptions of only five children. In every case, sufficient information is provided to indicate that the child would meet Criteria A and B as these would be operationalised nowadays, i.e., they would fail a standardised test of motor competence and concern was sufficiently voiced by parents and teachers for the child to be referred for a detailed medical opinion. From that point on, however, the situation becomes less clear and the problems presently faced with Criterion C are surely already foretold. For example, Walton's first case was initially referred to an orthopaedic surgeon due to limb pains and seems to have had weakness as evidenced by a query regarding a diagnosis of muscular dystrophy. Could this child's clumsiness have been symptomatic of benign joint hypermobility syndrome (BJHS), which typically features weakness, fidgetiness and nocturnal limb pain? The second case features foetal distress after a 5-day labour and forceps delivery at 43 weeks gestation. The authors comment that brain damage may have been an aetiological factor although there was no other evidence of brain damage in this boy with a verbal IQ of 113. Symptoms such as not walking until age two years combined with particular difficulty with articulation and even at age 14 to be

described as being virtually incapable of doing physical training, gardening, woodwork or art and with muscle action that remained clumsy, slow and ill-directed, in the context of the birth history is very suggestive of borderline CP.

In contrast to Case 2, the next two children in this series were both born prematurely but had slightly different birth histories. Whereas Case 3 was induced for pre-eclampsia one month early, Case 4 was born cyanosed, two months early. The former was reported as speaking phrases at age 15 months but following tonsillitis at 18 months did not speak easily again until aged around four years. Case 4, in spite of a verbal IQ of 105 was not using single words until two years and featured upper limb and oral dysfunction. These cases appear to be dissimilar both at an aetiological level and behavioural level.

Finally, Case 5 was born apparently without complication but had whooping cough at 12 weeks, pneumonia at 12 and 15 months leading to annual winter bronchitis. He did not walk until aged three years and at aged eight could neither hop nor jump. There was marked pseudoathetosis and abnormal signs were more noticeable in the left arm and leg. This child had a verbal IQ 87 and performance IQ of 44 opening up the possiblity that this child's problems were more widespread than those of the other four.

In summary, this study was one of the first to draw attention to children who would now meet DSM-IV Criteria i.e., with clumsiness "sufficient to interfere seriously with many motor activities essential to everyday life" (Criterion A/B). Yet "no defect in pyramidal, extrapyramidal or cerebellar pathways which control voluntary motor activity" (Criterion C). "All were of average or above average IQ but all demonstrating a lower performance than verbal score on formal I.Q. testing". (Criterion D). The syndrome as portrayed by Walton, however, is not precise about Criterion C and seems to admit of a much wider group of children than might be allowed today. As with DSM, Walton makes it clear from the beginning that **homogeneity of aetiology** is <u>not</u> a defining feature.

The second of the early papers that is of relevance to this debate is by Prechtl and Stemmer (1962) who studied a much larger group of 50 children aged 9-12 years, described as clumsy or awkward, but brought to notice on account of other concerns

too such as behaviour (poor concentration) or school-work (90% had reading difficulty). According to Prechtl & Stemmer, these children demonstrated various twitching movements, which were not considered to be tics or myoclonic spasms. Electro-myographic (EMG) recording, using skin electrodes, picked up discharges in muscles during contraction but also "in fully relaxed muscles in which there are no visible movements" (Prechtl & Stemmer, 1962, p. 120) and commented that "Choreiform activity is most clearly observable in stress situations". It was frequently observed in the upper body especially eye muscles. Muscle tone was reported as normal in 76% of the children, increased in 14% and reduced or hypotonic in 10%. Prechtl and Stemmer do not cite Walton and it is therefore not clear whether they were aware of each others terminology or whether they felt the clumsiness they described deserved a different label to clumsy child syndrome. Interestingly, later in his career, Prechtl became much more famous for his assessment of distinct movement patterns (including tremors and twitches) in preterm, newborns and young infants predictive of cerebral palsy and developmental deficits in later life (Einspieler et al., 2005).

Consideration of how Prechtl and Stemer's children fared in relation to DSM criteria, confirms once again that criteria A, B and D are relatively uncontroversial with C proving more problematic. Prechtl and Stemmer explicitly state that they excluded from the sample any child with obvious neurological signs, psychiatric symptoms, and extensive laboratory tests were included to screen for rheumatism and toxoplasmosis. However, 42 percent of the cases had pregnancy complications and 46% had neonatal complications. Postnatal history included 12% who had frequent epileptic attacks and 38% with a history of concussion. On this basis, Prechtl and Stemmer suggest that the choreiform syndrome that they describe, results from injury to the infants nervous system by pre- para- or post-natal complications. They also refer to hypotonia and to mild cerebral palsy and to symptoms being exacerbated by stress. There are suggestions of children's clumsiness that may be particularly influenced by environmental demands on internal regulation and adaptability, stresses emanating either from within or out-with the body. Interestingly, the Prechtl and Stemmer study is one of the first to explicitly list difficulties associated with clumsiness. The mention of associated reading problems, hyperactivity and outbursts of aggression hints at the question of 'co-morbidity' that so many are struggling with 40 years on (see Chapter 3).

To summarise again, these two studies, along with the others summarised in the BMJ article served a very important function. Unfortunate as it might seem nowadays, the use of the label 'Clumsy Child' with a capital C was a milestone on the road to recognising that milder motor problems as opposed to the severe movement problems of the cerebral palsies (a) might not be rare and (b) could have long lasting effects on a child's life. The difficulties listed in these early descriptive studies laid the foundation for the main features of DCD currently included in DSM and ICD.

Between 1962 and the present day, the number of publications on 'clumsiness' in children has grown exponentially. The Oxford 1962 meeting which brought not only Walton, Prechtl, Illingworth, Hagberg, and Paine together but also included many hugely influential voices in this field – Bax, (1999) Gordon (Gordon & McKinley), 1980), Fog (Fog & Fog, 1963), Ingram, (1984), Mackeith, (1968) to name just a few led later to a spate of international meetings on DCD, the most recent of which was attended by over 160 delegates (DCD VI, 2005). Linked to these meetings were special issues of journals as diverse as Adapted Physical Activity Quarterly (Henderson, 1994), Human Movement Science (Barnett et al., 1998) Neural Plasticity (Hadder-Algra & Gramsbergen 2003) and Child Care Health and Development (Zoia et al., 2006). In parallel with the increase in journal articles, the number of books on the topic also continues to grow (Reuben & Bakwin, 1968; Gubbay, 1975; Arnheim, & Sinclair, 1975; Gordon & McKinley, 1980; Cratty, 1994; Sugden & Wright, 1998; Missiuna, 2001; Cermak & Larkin, 2002; Sugden, & Chambers 2003; 2005; Dewey & Tupper, 2004).

In the UK, a series of studies begun by Henderson and colleagues (Henderson and Hall, 1982; Henderson, 1987; 1992, 1994; Losse et al., 1991) continued to carry the torch that Walton and colleagues had lit, and played an important role in overturning the view that there was no need to worry about these children as they would 'simply grow out of it'. Further confirmation, through longitudinal studies, of this view came from studies by Cantell et al., 1994, 2002; Geuze and Borger, 1993; Swedish studies were carried out by Gillberg and colleagues (Gillberg et al., 1983; Gillberg & Gillberg, 1988; 1989, Hellgren et al., 1993; 1994, Rasmussen & Gillberg, 2000), some of which followed children from age seven to 20 years, longer than Losse et al. (1991), and showed that many continue to suffer into adulthood in various ways.

Research from Lancaster, UK similarly indicated that problems continue into adult hood (Cousins & Smyth, 2003). Geuze et al. (2001), provide a succinct summary of this research, being careful to note that lack of comparability continues to be a problem. They note, for example, that most authors assume 'normal intelligence' and do not venture into the murky waters of clumsiness in less able children. However what is convincingly apparent is that none suggest that a syndrome with motor difficulties as its core does not exist. For the moment, therefore, we leave all the problems associated with co-ccurring difficulties aside and proceed as if the syndrome, now labelled DCD exists.

2.3.5 Clumsiness as a condition with clearly definable sub-types

When discussing definitions earlier in this chapter, a few points about the concept of a syndrome with subtypes within it were made. One of the most important concerned the validation of a subtype, how this might be done and why it was crucial to do so. As Rutter (1998) points out, the identification of one or more subtypes within a syndrome only makes sense if, in the ultimate analysis, this leads to more refined treatment for the condition as a whole. Sometimes, of course, this process takes a long time and the existence of systematic subtypes may be known and understood before differentiated treatment is tested. An example is Parkinsonism, a syndrome which resembles idiopathic Parkinson's Disease (PD) which was identified in late 19^{th} century. The symptomatic motor problems vary between primarily tremor and shaking of the body or predominantly loss of movement and rigidity. In recent years the sites for stereotaxic neurosurgery have become extremely precise, directed at ablation or stimulation of an area/s of the brain, related to different signs. In childhood disorders a sub-type of cerebral palsy which features spasticity is now frequently treated with injections of Botulinum toxin whereas this would usually be contraindicated in the pure athetoid sub-type.

Quest for subtypes within a syndrome does not necessarily involve concentration on searching for clues at the aetiological level. A rather different approach starts at the surface level, asking whether the motor behaviour itself may present in different ways. A good analogy may be made with language or reading difficulties in children. Irrespective of aetiology there are characteristic differences in the symptoms displayed. For instance, within developmental language disorders, a distinction can be drawn between receptive and expressive language problems. In the area of reading

difficulties, a phonological deficit can be distinguished from a visual perceptual problem, and this in turn leads to a completely different approach to remediation/ intervention. In the area of DCD, we are a very long way from identifying and validating meaningful subtypes.

When one looks at the studies reviewed above with this question in mind, several hint at the possibility of subtypes. For example, the possibility that some children within the DCD umbrella have a mild form of CP has already been raised. Proponents of this view have nearly always come from a medical background and have used neurodevelopmental tests to draw their conclusions. Using a variety of hard and soft sign batteries, children have been divided into those who do and do not meet criteria for neurological impairment. An example of this approach can be found in the work of Hadders-Algra and colleagues (Hadders-Algra et al., 1988; Hadders-Algra & Gramsbergen, 2003), who use a test designed by Touwen (1979) to distinguish between 'simple' and 'complex' neurological dysfunction, arguing that the prognosis for these two groups of children is different. Although these studies are relevant to this debate, however, none were actually designed to test the idea of subtypes per se. Morever, none has been subjected to any of the validation techniques suggested by Rutter and others.

There have been various other attempts to look for subtypes within the broad category of children one might label DCD, some of which are linked to professions other than doctors. One of the best examples of this has its origins in occupational therapy and has as the over-riding concept, 'Sensory Integrative Dysfunction'.

Put briefly, Ayres theoretical position places particular emphasis on the importance of the near senses (tactile, vestibular/proprioceptive) in underpinning the development of human skills (Ayres, 1965; 1972; 1980; 1985; 1989; O'Brian et al., 1988). In addition, she has much to say about various sensory-motor deficits and planning deficits, which she believed can be identified in children using her tests. In practice, the application of the Sensory Integration and Praxis Tests (SIPT) produces a profile, which is then computer-matched to measure how closely the child's perceptual-motor profile fits one or more identified SI sub-types. Sub-types have included the following:

- Sensory modulation disorders: the child has difficulty regulating arousal (sensory seeking or avoidance or swinging between the two extremes).
- Somatodyspraxia: the child's planning difficulty is deemed to be related to an underlying tactile discrimination dysfunction. Children especially demonstrate problems in learning new tasks and disordered tactile and kinaesthetic processes affecting knowledge of body postures and grading of muscle action.
- Bilateral integration and sequencing deficits: here the child's dyspraxia is thought to be related to a vestibular-proprioceptive problem. This profile features difficulty in coordinating and sequencing actions across the two halves of the body.
- Dyspraxia on verbal command: linked to localised left cortical dysfunction.
- Visuodyspraxia: linked to localised right cortical dysfunction.

At present in the SI field the trend has been to collapse and simplify the profiles into either modulation and/or dyspraxia sub-types. The important point is that for each of the sub-types identified by the SIPT battery the Ayres approach recommends different intervention techniques. At present, however, empirical support for these subtypes is lacking.

Whereas tight ring fencing of SI within OT has stifled the potential contribution of Ayres' work, researchers from different professional backgrounds have been pursuing other hypotheses about the underlying causes of clumsiness in a more accessible manner. For example, researchers, from a variety of disciplines including experimental psychology, human movement science, medicine and physiology have examined deficits they believe underlie the clumsy phenomena from every aspect: visual perceptual processing (e.g., Lord & Hulme, 1988; Rösblad & von Hofsten, 1994), kinaesthetic perceptual processes (e.g., Laszlo & Bairstow, 1983; Sims et al., 1996; Coleman et al., 2001), vestibular function, (Horak et al., 1988) deficits of postural control (Williams et al., 1983; Wann et al., 1998) and problems with speed of processing (Geuze & Kalverboer, 1987) - to name just a few. Although many of these studies are well designed and have produced quite robust results, like the medical studies, they were not designed to identify subtypes within DCD, and rarely even mention individual differences between the subjects. What this chapter now turns to, therefore, is a group of studies which address the question of subtypes directly using a technique known as cluster analysis.

Cluster analysis is a general name given to a collection of statistical techniques that may be used to arrange a set of objects (e.g., individuals) into groups or clusters. Between 1992 and the present day there have been six studies which have used this technique in the area of DCD (Jongmans, 1993; Dewey & Kaplan 1992; 1994; Hoare, 1994; Miyahara, 1994; Wright & Sugden, 1996a; Macnab et al., 2001). There have been two recent reviews of these studies, each dealing with a different subset (Macnab et al., 2001; Visser, 2003). Both call into question their comparability.

Briefly, the problems begin with the simple variable, age. In total, over 500 children aged between six and 14 years took part in these investigations. At one end of the spectrum there is a very focussed study, like Jongman's which included 90 children all of the same age, i.e., six years. At the other end, there is the study by Miyahara, which has a larger number of subjects but a much wider age range, extending from six to 14, i.e., into adolescence, a period of major physiological changes, which have a bearing on muscle function and motor performance. In between, there are four studies, in which subjects vary in age from six to 11 years. While this in itself makes comparison difficult, the age factor is minor in comparison to other differences.

The next problem concerns the procedures used to obtain the samples, and the extent to which the children included in the studies could be said to meet DSM criteria. Although all six studies were completed following publication of DSM III (American Psychiatric Association, 1987), it is not at all clear which groups of children would actually meet all four criteria. Of the six studies, Jongman's stands out as exemplary in the extent to which selection criteria and assessment procedures were specified. For example, it is very clear how the children were selected for the study, which tests were used, and a great deal about the children's developmental history. In contrast, the study by Hoare (1994) describes how the children were identified in reasonable detail, but no information on IQ is available, and no medical screening of any kind was undertaken. Consequently, one has to assume that because the children attended mainstream schools, they were of at least average IQ. Similarly, the Dewey & Kaplan (1994) study is reasonably clear on the methods of identification and assessment but no data that would permit one to apply Criterion C are offered. With regard to Criterion D, Miyahara's study stands out as the least comparable to the other five. Whereas he drew his cohort from a USA private school for children with

known learning difficulty and his comparison group was described as 'less learning disabled', all of the other studies involved children from mainstream state schools, albeit in different countries.

Not only were the samples drawn from rather varied sources but they were also identified by methods specific to different health and educational professionals. For example, the children in the Hoare (1994) study, were referred through a movement education programme in Australia. Confirmation of DCD, made by education professionals, was based on performance of the McCarron Assessment of Neuromuscular Development (MAND; McCarron, 1982). Scores on the 10 items of the MAND are converted to a Neuro-Developmental Index (NDI). Hoare included children who scored < 90 on the NDI although the 1SD cut point is 85 (i.e., the children had to fail only 40% of the NDI items to be included). In contrast, Dewey and Kaplan (1994) recruited from a Canadian public school and classroom teachers initially identified children with motor skills problems. These children were then screened by an OT using four items from the Southern Californian Sensory Integration Test (SCSIT; Ayres, 1980) plus a vestibular item. Children included, scored at least 1SD below the mean on one of the measures. Macnab et al. (2001), used a combination of teachers selecting children with movement difficulty and a 'qualified clinician' then identifying those children who fell at least 1SD below the mean on the Bruininks-Oseretsky Test of Motor Proficiency (BOTMP) (Bruininks, 1978) and Test of Motor Impairment (TOMI, Stott et al., 1984).

In addition to lack of comparability of the samples in these studies, the differences in variables entered into the cluster analyses poses even more difficulty. Two examples must suffice to illustrate this point. First, all six studies included a standardised norm-referenced assessment of some sort, but not the same one. For instance, the BOTMP, was used by Dewey & Kaplan (1994) and by Miyahara (1994), and M-ABC, used by Jongmans (1993) and by Wright & Sugden (1996a). While these tests have some items in common the correlation between them is not very high. Moreover, whereas some studies e.g., Jongmans used the individual item scores in their cluster analysis, Hoare used subtotal scores. Second, one of the most important differences lies in the 'level' of item entered into the analysis. Whereas Wright and Sugden's (1996a) study included a standardised teacher checklist with emphasis on function in a natural environment, Hoare (1994) and the replication of her study by

Macnab et al. (2001) used measures of underlying processes such as kinaesthesis. Finally, it might be useful to comment on a few of the many variables that could have been but were **not** included in these studies. In the field of research into DCD anthropomorphic measures such as musculo-skeletal elasticity or flexibility, body mass index, height span index are rarely considered. Yet, these are known to influence motor development particularly around puberty when gender differences especially in relation to onset, muscle strength, flexibility, body shape and proportions become most obvious. In an interesting study, physical growth and activity level were shown to affect typically developing children and children with DCD differently with some of the DCD group appearing to profit from the growth spurt Visser et al. (1998). The participants included in the cluster studies were all selected by a 'motor difficulty' criterion however only one of the studies provides data on co-existing conditions, an issue which will be discussed later.

In view of the fact that it is so difficult to compare these six studies, one might ask whether there is any point in examining the outcome of the cluster analyses at all. In spite of all of the difficulties, however, there were some similarities in outcome. The studies produced rather similar numbers of cluster groupings (4-6) (see Table 2.4). A researcher uses post hoc discriminant analysis to determine and verify the most appropriate number of cluster groups by the percentage of correct predictions. High percentages were achieved in all the studies. In all cases, a group of children who scored poorly on everything, compared to the group mean is reported. That there is usually a 'poor' performing group may point to a subtype of children who feature more pervasive functional difficulty linked to a greater degree and/or wider spectrum of dysfunction. Certainly in the Hoare study (1994) on the neurodevelopmental index (NDI), 64% fell into the mildly disabled group, 31% moderately disabled and 5% severely disabled. This last group, were poor across all variables and were labelled 'learning disabled' by their teachers. One might ask whether this group was more neurologically impaired or whether any children within it fell into a borderline CP sub- group? Without a detailed health history this cannot be reliably ascertained. For a possible answer to these questions, however, it might be useful to turn to Jongmans's study which also identified a similar poorly performing group ('cluster group 2'). When she examined other aspects of her group of children who were poor on all of her motor measures, she found that their short-form IQ scores were lower, that teacher behaviour ratings of members of this group indicated that they were

likely to experience associated problems and most importantly, every child in this cluster had suffered a brain lesion in the neonatal period.

In addition to the fact that all six studies found one group of children who were 'poor at everything' and a trend toward a group who were around average regardless of the variation in variables entered into the analyses, there were hints of possibly interesting dissociations between the functions/processes measured. Dissociation in this context refers to separation due to unevenness of scores. There was some consistency in that studies reported a group with poor balance. Clinical observation would suggest one might find a group of children with good manual dexterity skills and poor balance or the converse. Macnab et al. (2001) identified one group with good visual perception and poor gross motor function and another group with poor fine motor and visual perception but average balance and kinaesthesis. Jongmans (1993) noted one group who were comparatively poorer on static balance than on dynamic balance and a different cluster which featured good fine motor skills but poor balance. Miyahara (1994), as noted above, recruited from a different population with additional learning difficulty. However he also showed a differential on the balance task in two of his cluster groups. In summary, although some interesting profiles are identified from the cluster solutions produced statistically, the studies do not show any consistent sub-types and only in the Jongmans study is any attempt to validate the subtypes undertaken.

Table 2.4 *Cluster Studies: summary of main features*

Author Year	Sample Source	N Age (Years)	Identification Screening	Assessment	Variables	Clusters	Main features of each cluster
Jongmans 1993	One UK Hospital	90 pre-term 6 years	All pre-term. Identified by health professionals	Man.dexterity. Ball skills. Stat. Balance. Dyn. Balance Construction.	M-ABC VMI DAP Form board	6	1 Average on all variables 2 Poor on all except manual dexterity coin- posting 3 Poor balance, good fine motor 4 Poor static balance but good dynamic balance 5 Poor ball skills 6 Poor construction ability
Dewey & Kaplan 1994	Public Schools Canada IQ > 70 (as reported by school)	51'DCD' 51 'normal' 6-11	School teachers.	Experimental group only screened by OT (incl. If fail on 1 SCSIT item).	2 hour OT asses: BOTMP Gesture. Motor sequence. Balance.	4	1 Poor on all measures 2 Poor balance, coordination, transitive gesture 3 Poor motor sequencing 4 Above average on all measures
Hoare 1994	Movement Education Programme. Australia. Regular School.	79 'DCD' 6-9	Teachers. 'Neuro-developmental index' from 10 items of MAND No medical screen.	PE specialists. Purdue peg Board. Kinaesthetic Acuity Test. VMI. MVPT. Static Balance. 50 yd. Dash.	Kinaesthesis Vis. Percept Vis. m. Fine m. Static bal. Running	5	1 Poor running and kinaesthesis. Good static balance. 2 Average in all areas. Good visual judgement. 3 Poor across all variables. Considered 'LD' by teachers. 4 Kinaesthesis & run good. Visual processing poorer. 5 Poor dexterity, balance & run. Kinaesthesis above average.

....

Table 2.4 *Cluster Studies: summary of main features (cont.)*

Author Year	Sample Source	N Age (Years)	Identification Screening	Assessment	Variables	Clusters	Main features of each cluster
Miyahara 1994	Private school for Learning Difficulty (LD approx -2 yrs). USA. IQ $\geq$ 70	55 'LD/DCD' 90 'less LD' 8-14	PE teachers.	Certified PE teachers trained PE teachers to use BOTMP	Running. Balance. Bilat. Coord. Strength. Upper Limb Coord.	4	1 Free from motor problems. 2 Poor across all variables. 3 Good balance. Poor ball skills, running, strength.. 4 Poor balance. Good strength.
Wright & Sugden 1996[a]	Mainstream School Singapore	69 'DCD' 6-9	PE teachers No health screen	Open skills Fine m. Catching. Dyn. Balance. Control self.	M-ABC	4	1 Borderline DCD 2 Poor catching 3 Poor 'control of self' forward plans ('Open skills') 4 Poor peg board & dynamic balance
Macnab et al. 2001	Canada	62 7-12	Referred by teachers to school health. No medical screening.		Kinaesthesis Vis. Percept Vis. m. Fine m. Static bal. Running	5	1 Good balance 2 Good visual motor and dexterity. Poor kinaesthesis. 3 Poor all areas (including visual and kinaesthesis). 4 Poor fine motor, visual motor and visual perception. 5 Poor gross motor. Good visual motor and vis. percept.

2.4 Conclusion

At the beginning of this chapter, two basic questions were addressed. The first was – 'Does DCD exist'? and the second was 'If it exists, what is the evidence for subtypes within the syndrome?' On balance, the answer to the first question must be 'yes'. The overwhelming evidence of research studies over many years supports the existence of a cluster of core movement symptoms, regularly occurring together, and labelled as the 'clumsy child syndrome' or subsequently DCD. These children are distinguishable from children with severe neurological impairment such as CP or DMD and apart from their clumsiness many function similarly to their typically developing peers. However there is a grey area where it is harder to be definite as to where the boundary between DCD and neurological conditions falls. The syndrome, rather like malabsorption syndrome referred to previously, is a recognisable discreet syndrome which is comprised of a collection of symptoms but which has many possible causes. It is heterogeneous and not one unitary syndrome with one cause and one clearly defined profile. This clumsy syndrome may overlap the very mild end of several conditions such as CP, musculo-skeletal problems and research in the future may clarify the present 'fuzzy' boundaries around these conditions and possibly define subtypes. It is therefore perhaps helpful to look at comments made in relation to CP, to see if there is anything we can learn.

Badawi et al. (1998) addressed the problem of standardizing the inclusion criteria for cerebral palsy registers. We can draw a parallel with her title 'What constitutes cerebral palsy?' by asking: what constitutes a syndrome of DCD? Badawi states "Cerebral palsy is a term of convenience applied to a group of motor disorders of central origin defined by clinical description. It is not a diagnosis in that its application infers nothing about pathology, aetiology, or prognosis. It is an umbrella term covering a wide range of cerebral disorders which result in childhood motor impairment. ....With the advent of improved diagnostic tests such as chromosomal analyses, metabolic studies, and new imaging techniques it has been possible to identify previously unknown causes in children classified as having CP" (Badawi 1998, p. 526). Similarly we should ask whether children presenting with less visible motor disorders (DCD) who may over time meet criteria for a variety of newly recognised diagnostic labels (e.g., chromosome abnormalities) should be included or excluded from classification within a DCD category. Badawi concludes that, although the concept of CP as a separate entity may "be outdated for aetiological

objectives", the separation of motor impairment "remains useful for service provision and management." She appends her paper with 100 syndromes associated with motor impairment. The dividing line between the motor impairment of frank CP and the movement difficulties synonymous with clumsiness is a grey area. The documented research into sub-typing of CP is both relevant and informative to the present issues around DCD as a symptom or syndrome and the search toward development of appropriate specific interventions.

Similarly one might put forward that Developmental Coordination Disorder (DCD) might more appropriately be termed the Developmental Coordination Disorders with a variety of causes and presentations. The compounding effect of social and biological interacting factors on aetiological processes underlying DCD should always be taken into account. There are also parallel difficulties to those itemised for CP in terms of classification of the disparate guises or faces of DCD, which may overlap the cerebral palsies but may also overlap or co-exist with other developmental disorders. The concept of DCD as one of many related developmental syndromes or one face of a more general atypical development will be the focus of the next chapter of this thesis.

Chapter 3

Syndrome or Syndromes: DCD Plus or What?

3.0 Introduction

In the previous chapter, the focus of attention was on the **motor** problems experienced by a small proportion of intelligent children, who do not suffer from any (known) physical disease or disorder. The primary problem addressed was whether there is enough evidence to support the idea that the motor difficulties these children exhibit, constitute a distinct syndrome, which 'stands alone and can be clearly and reliably differentiated from other childhood disorders'. Although not exhaustive, the review of relevant literature described many of the medical conditions that feature movement difficulty or apparent 'clumsiness' as one symptom. By pointing to some of the similarities between these conditions and DCD, this review stressed that (i) separating some medical conditions from DCD is not a simple matter, and (ii) the dividing line between typical and atypical motor development is not as clear as might be imagined. After considering data and arguments in favour of the idea of a discreet syndrome, however, it was concluded that the concept of DCD as 'a cluster of symptoms amounting to an identifiable idiopathic syndrome with undoubtedly heterogeneous causation' might well be viable. Rather than conceptualise DCD as a unitary syndrome, however, there is an alternative idea currently receiving much attention in the literature. Basically, this throws out the concept of DCD as one of many (albeit) related developmental syndromes, and proposes that **all** developmental disorders can be viewed as one variable phenotype expression reflecting unusual development of the central nervous system.

In DCD, as in many instances in medicine, pure cases are the exception rather than the rule (Hill et al.,1998). Although several of the longitudinal studies mentioned earlier describe some children with DCD whose movement difficulty was an isolated phenomenon (Henderson & Hall, 1982), it is far more common to find children whose 'clumsiness' occurs alongside difficulties in other domains of behaviour. These may include reading, writing, speech and language difficulties, distractibility, hyperactivity, problems with social interaction, conduct problems and poor self esteem (e.g., Losse et al., 1991; Powell & Bishop, 1992; Kaplan et al., 1998; Dewey

et al., 2002). How to deal with phenomena apparent across several domains of function is the problem that is addressed in this chapter.

3.1 Concepts and definitions

In the previous chapter, the disagreements over the meaning of the terms such as sign and symptom, syndrome and disease, were seen as largely unresolved in many instances. Terms which are used to describe two conditions or problems which occur in one individual at the same point in time provoke equally heated debate. Table 3.1 shows a selection of such terms, along with their dictionary definitions. From this table, it is immediately apparent that most of the terms can be viewed as synonyms except perhaps for co-morbid which is given a specific medical meaning. As with other discussions of terminology in this thesis, however, what one finds here is that usage and interpretation of terms is in part dependent on context, including professional allegiances.

Table 3.1 *Definitions of Association, Concomitant, Co-occur, Co-morbid, Overlap, in alphabetical order*

Association: "A conceptual connection"
Co-exist: "Exist together"
Concomitant: "Accompanying"
Co-morbid (medicine): "Relating to or denoting a medical condition that co-occurs with another"
Co-occur: "Occur together or simultaneously"
Interrelate: "Place or come into mutual or reciprocal relationship"
Overlap: "Partly coincide in time"
Be associated with: "A concept connected with another (chiefly psychology)
Related: "Accompanying; allied, associated, connected, concomitant, connected, linked"
(Concise OED, 2004)

In general medicine, it is accepted that most elderly men will have evidence of osteoarthrosis and malignant cells in the prostate. These conditions co-exist but no one assumes that they are related. In contrast, a person may have a stroke resulting in paralysis of the right side of the body (hemiplegia) with concomitant aphasia. The speech difficulty is not a separate condition but is caused by and therefore related to

a common underlying pathological process. A different example might be a person who has a hemiplegia resulting from a cerebral tumour which is a secondary growth related to a primary cancer of the bronchus. The primary cancer may or may not overlap or co-occur in time but is associated with it. The same person may have co-existing smoking-related chronic bronchitis. In each case, the relationship between the various symptoms/diseases described might be complex but their relationship is in the end, clear.

In developmental disabilities, however, the situation is more nebulous. In young children, DCD may be present alongside specific language impairment (SLI) and the two conditions may overlap temporarily or permanently. They may sometimes be presumed to stem from the same aetiology such as damage to the developing nervous system resulting from a brain lesion associated with premature birth. At other times, no clues to aetiology are available, however hard the clinician tries to find them. Does this make DCD and SLI one single condition or two? As the defining features of DCD i.e., 'clumsiness' of movement, and of SLI - 'expressive or receptive communication difficulty', are also defining criteria for Asperger Syndrome are these three conditions comorbid, co-ocurring or overlapping?

Terms such as co-exist or co-occur, which are comparatively 'neutral', do not on the whole raise difficulties. They simply imply a temporal relationship and nothing more. Similarly, the word associated is fairly uncontentious, suggesting two or more conditions that may occur together and may indeed be related at some level but do not necessarily overlap in time. In contrast, considerable confusion arises over the term co-morbid. On the one hand, some authors suggest that the research community should stop using the word on account of it denoting closer identified medical links than are found in most childhood developmental disorders (Kaplan et al., 2005). On the other hand, there are authors such as Angold and colleagues who take a more positive view (Angold et al., 1999). Within child and adolescent psychiatry, a whole range of specific diagnoses with separate entries in DSM-IV are identified. Angold et al. (1999) are content to use the word comordidity, and see its presence not so much as an embarrassment to categorical diagnosis but rather as providing an opportunity for better understanding of psychopathology. Angold et al. (1999) reviewed research on the prevalence, causes and effects of diagnostic comorbidity within this field of enquiry, including disorders such as Attention Deficit Hyperactivity Disorder

(ADHD), anxiety and conduct disorders. Like Kaplan, they emphasise that in medicine, comorbidity, relates to known **diseases** rather than behavioural and psychological **syndromes** in children, which may be much less well established than disease categories. However, they comment that explicit attention to comorbidity as judged by citations in PsycINFO has increased exponentially since 1986 and reiterate a view by Kendall and Clarkin (1992, p. 833) that the study of comorbidity is the "premier challenge facing mental health professionals in the1990s".

In their review, Angold et al. (1999) try to untangle different types of comorbidity. They underline a difference in the time line of a disorder, referring to homotypic and heterotypic continuity. In the context of DCD, the former might apply to a child who is 'clumsy' at seven years and displays on-gong motor incordination as an adult. Heterotypic might apply to a child who was 'clumsy' at seven years but by adulthood the motor feature was no longer present but s/he presented with a behavioural or emotional disorder – heterotypic comorbidity. Concurrent versus successive comorbidity they suggest covers a multitude of temporal relationships amongst disorders. They further point to familial comorbidity where rates of various psychological disorders are higher within families for instance where a child has ADHD. Finally, there is the problem of 'epiphenomenal comorbidity' where three conditions appear associated but one pair-wise association may simply be due to chance, arising from the probability of an association between the other two pairs.

In summary, Angold and colleagues conclude that comorbidity is unquestionably 'real', while at the same time conceding that some of the problems it appears to create may arise because the diagnostic system draws inappropriate boundaries between disorders. They emphasise the need to clarify diagnostic boundaries and further describe correlates between 'pure' and comorbid disorders as they appear and disappear over time. They stress the importance of caution in equating general population samples with clinic-referred children as these may differ especially in complexity and severity. With regard to understanding DCD and how it relates to other disorders, all of the points made above are relevant. At the moment, however, the amount of empirical data addressing the issues is limited, placing professionals and researchers in the field of motor disorders at a disadvantage.

3.2 Co-occurring, co-existing, co-morbid from the client's perspective

Chapter two discussed briefly the sorts of concerns that families and schools have regarding a child with DCD. Whereas the primary focus was on the motor features that parents and teachers notice, also touched on were symptoms such as speech difficulties or unusual behaviours which might mask the movement problem. In this section the co-occurring symptoms are given prominence.

Consider the child described in Chapter 2, who displays tantrums, that parents find so difficult to cope with that they urgently seek advice on management of the disruptive behaviour rather than any co-existing motor symptoms. The 'little professor' mentioned in the same section impresses with his knowledge. Consequently, his lack of social interaction at playtime which may co-exist alongside weak motor skills, may either not be recognised at all or be rated as of little importance.

Parents often report problems in several domains of function. They may first notice that a child seems much more hyperactive than friends. The child is described as 'a bull in a china shop' restless and never sitting still, climbing onto furniture with little heed of danger. The same child may also accidentally break toys by clutching them too tightly or inadvertently dropping them. When running, parents notice that the child trips over every obstacle and in addition flies into a temper when thwarted. At school, the teachers find that the child constantly has to be reminded to try not to fidget and to listen and attend to instructions. Rather than settle down to writing the child flits from one distraction to another, chatting with a slight stammer to neighbours, yet at times contributing creative information to class discussion. Writing is untidy, letters are often reversed and words mis-spelt. Although appearing to be one of the brightest members of the class the child just does not seem to catch on to reading. Above all, the parents and teacher are concerned because the child is frequently argumentative, uncooperative and quite naughty. This child may display DCD but it is obvious that there are symptoms in several other domains suggestive of AD/HD, SLI, Dyslexia and perhaps ODD (oppositional defiant disorder).

Another, rather different example might be a child who has difficulty with interpersonal communication and seems rather aloof to his friends. Parents of this

child note that, although certainly a bit clumsy and reluctant to use a knife and fork, the child stands out because of an inability to cope with any change in routine. When they or the teacher suggest doing a task in a different way or trying new activities the child resists and seems only interested in playing, often alone, with the same toys laid out in identical order. This child, rather than generally tending to fly into a temper, becomes cross and agitated at very specific times, for instance when asked to use a pen or catch a ball of a certain colour or texture. Again there is no doubt about ability, and both parents and teachers are impressed by the child's wealth of information on a few very specific topics. Here, the child's symptoms suggest a syndrome of motor difficulty (DCD) co-occurring with (or part of, as some would suggest) Asperger's syndrome or OCD (obsessive compulsive disorder). The symptoms of motor difficulty and disordered communication overlap DCD and SLI. In terms of Angold et al. (1999), these two examples illustrate heterotypic comorbidity. In contrast, parents may remark that at first a child received a diagnosis of language delay (SLI) and met with a speech and language therapist. A few years later, at primary school, language was not a problem but the child presented with some motor difficulties and attention deficit and was hyperactive and underachieved. Symptoms calmed to general restlessness by school leaving age and the youngster achieved further education and a series of jobs but subsequently turned to alcohol and drug-use which seemed to calm the restlessness. Here Angold's framework of concurrent and successive comorbidity underlines the fact that neither diagnosis nor comorbidity are necessarily static.

3.3 Co-occurring, co-existing – the practitioner's role

The previous chapter highlighted the many different developmental conditions which have a distinct physical dimension that may present with a common symptom of 'clumsiness'. The main focus in this instance, therefore, was how the various professionals with knowledge of this area of paediatrics could coordinate the process of differential diagnosis. Although this process can be complicated enough in itself, the question of how different professions deal with all the co-existing problems just mentioned is even more complex. Of particular concern, in this thesis, of course, is how the movement problems that children experience are recognised and treated appropriately.

In previous sections, it was noted that the picture a child presents is frequently not static but can change over time, with different elements of his/her problems seeming to vary in severity. Just as parents may focus on only one aspect of a child's difficulties at any one point in time (e.g., movement, speech and language, behaviour or learning), so too do professionals. Early speech and language difficulty may mean that a speech and language specialist is the appropriate key person who provides intervention at this point in time and the focus is rightly directed quite specifically with a positive outcome. Focus on just one domain, however, can on other occasions be disastrous. For example a child may demonstrate difficulty in social interaction and meet primarily with clinical psychologists or psychiatrists. The child may have concurrent motor problems, which prevent him joining in with friends in the playground and he may also be faced with a struggle to produce handwriting or fasten his buttons. Although function in the social domain is essential for good peer relationships, motor skills are also the foundation, and provide a vehicle for developing relationships and acceptability in the school playground and progressing academically, so that to ignore motor problems is detrimental.

Similarly the difficulties that a child experiences in one area at one point in time, may be sufficiently severe to warrant a label, which highlights a domain, other than the motor aspect. Thus the child may be diagnosed as having speech and language impairment (SLI), reading disorder (RD) (dyslexia), attention deficit disorder/hyperactivity disorder (AD/HD) or a pervasive developmental disorder (PDD) such as Asperger syndrome (AS). In such cases, good intervention may be provided for the domain highlighted by the label. However, if associated motor difficulties (which may in some cases be the major underlying problem affecting function) are ignored, the kernel of the disorder may not be addressed.

Another aspect that may lead to variability and inequality once again relates to service provision by professionals and may even reflect geographical variation. The label any one child is given may depend upon the professional doorway through which the child first enters the diagnostic process. So, for example, one area may have a strong psychiatric team to whom a child with AS or ADHD may automatically be referred. In another locality no such service is available and it is a lottery whether the child arrives at the threshold of speech and language therapy on account of SLI, at the door of the physiotherapist for assessment of 'clumsiness',

through the entrance to the occupational therapy department because activities of daily living are problematic or to a paediatrician for prescription of medication to calm hyperactivity. Again, the situation is not static, changes over time and varies with local protocols. A child may continue to attend physiotherapy long after the motor problem has largely resolved or perhaps remain within the occupational therapy service because therapy directed toward 'childhood occupation' is deemed to cover the entire breadth of problems. Where a child is known to be receiving professional intervention there is less incentive for parents or others to seek alternative advice or additional help. Whether it is the best intervention is not often questioned, or related to evidence of improvement, but it may be preferable for a different professional to concentrate on a problem in a different domain. For example, a child who tended to trip up and have poor postural control may subsequently need less physiotherapy intervention but more focus on his or her reading difficulty. A child attending cognitive behavioural therapy for attention difficulty may well fare better by preceding or complementing such input with physiotherapy aimed at strengthening the muscles for postural control to help provide stability for looking and listening.

Assigning a particular label may depend not only on presenting symptoms and which professional discipline the child reaches but also differences in the way individual professionals conceptualise neurodevelopmental conditions. Is the 'clumsy' child perceived as a medical, an educational or a psychological problem? In each of these broad fields motor problems may be weighted differently. From an educational perspective when children present with clumsiness but also learning, communication and socialising problems, the latter are often of greater import for the teacher. In the field of psychology the motor aspect may be seen as a minor feature alongside attention, emotional and behavioural difficulties.

A physiotherapist may assess the child mentioned in Chapter 2 who frequently trips up and tumbles but may not pick up on his delayed speech and language. The child with tantrums may meet with a psychiatrist who can provide behaviour modification but will often not address the child's coexisting motor difficulty. An essential role for the professional apropos DCD is to convince the wider world that problems in the motor domain matter. In a world fraught with comorbidity it becomes imperative that assessment should reflect the overlap or concurrent symptoms and diagnoses,

through multidisciplinary teamwork. The child labelled as DCD, ADHD, AS or SLI must ideally meet with a group of practitioners together (e.g., medical, health, education and psychology) each with specific skills. Alternatively, one professional may be involved as a key person but one who is aware of the different domains of dysfunction and can link with or refer on to colleagues.

Angold et al. (1999) cite comorbidty as a premier challenge to researchers in the present era. This is equally true for those involved in planning service provision. Practitioners are faced with increasing recognition of developmental disorders and are encouraged toward evidence based practice on the one hand yet constrained by limited resources and regulations on the other. Clinical practitioners and researchers, on the whole, function in separate worlds and their aims are different. Broadly, clinicians such as physiotherapists, focus on improving motor function in a child, as an individual, and only recently have they been required to demonstrate the efficacy of the treatment they offer to the world at large. Evidence-based practice and continuous professional development are now prerequisites for professional registration (CSP, 2005) so this has meant that practitioners can no longer ignore the research literature. Whereas researchers have long been familiar with ICD-10 and DSM-IV and have specifically adopted the term DCD, for example, many clinicians and teachers do not know DSM-IV and only recently has the diagnosis DCD become more widely used. So, what does the research literature have to say about how we should think about conditions such as DCD, SLI etc? Although strictly speaking not research documents, DSM and ICD purport to reflect current research and thinking about diagnosis and classification so the next section starts by re-examining the entries in these official manuals.

3.4 Developmental disorders – the status quo: separate entries in DSM and ICD

As noted above, both manuals start from the point that the various 'specific learning difficulties' can be separated one from the next. In spite of accepting the need for further debate, both manuals classify and distinguish problems in the domains of function mentioned previously, separately, but loosely grouped within Axis I 'clinical disorders', sub-headed 'clinical disorders usually first diagnosed in infancy, childhood or adolescence'. Thus, alongside DCD are several other specific disorders including learning disorders (reading, maths or written expression), communication

disorders (expressive and/or receptive language disorders). Other conditions placed adjacent in the section include Attention Deficit/hyperactivity Disorder (ADD/ADHD), Pervasive Developmental Disorders: Autism, Asperger Syndrome (AS); Tic and Tourette Disorders; Conduct Disorder (CD); Oppositional Defiant Disorder (ODD) and Obsessive Compulsive Disorder (OCD).

Each diagnostic entry in DSM-IV or ICD-10 begins with a definition and description of the essential or main features of a condition, which then comprise the main diagnostic criteria. A list of associated or secondary features and symptoms of disorders is also usually provided. What is most important, however, is that the essential criteria must be met in order for a diagnosis to be made whereas the associated features are often but not necessarily observed or present. Table 3.2 gives examples from DSM-IV of symptoms in the motor domain listed as comprising the essential (main) primary criteria and those, which are appended as associated secondary features. It is immediately apparent from the table that there are many common features and associations across disorders. For example, Attention-Deficit/Hyperactivity Disorder (ADHD) recurs prominently as an associated feature in other conditions e.g., language, tic and conduct disorders. Learning difficulty is mentioned under most of the disorders and in all the entries there is either a main or associated motor symptom.

As Table 3.2 shows 'clumsiness' is listed as the essential or main (primary) diagnostic feature in DCD and as an associated diagnosis/ feature of many disorders. In the case of DCD if one follows DSM-IV Criterion C, the label DCD would be quite clearly and confidently dismissed when a diagnosis of muscular dystrophy or severe autism is made. However, a major point of discussion in the literature today centres on how one should deal with the fringes or fuzzy edges at the boundaries between conditions or what Gillberg (2003) refers to as 'shadows'. A dictionary definition of 'shadow' is a 'slight trace' (also an 'inseparable companion'!) (Collins, 1995). Recognising the definition of the shadow depends on which features stand out visibly. This leads to the consideration of rating observable symptoms in developmental disorders along a dimensional primacy/hierarchy.

Table 3.2 *Examples of DSM-IV (2000) Disorders: Main and Associated Features*

Code. Disorder	Essential or Main Feature	Associated Features & Disorders
F82 Developmental Coordination Disorder (DCD)	"Marked impairment in the development of motor coordination"	"...delays in other nonmotor milestones. Associated disorders: Phonological D; Expressive Language D and Mixed Receptive-Expressive Language D.
F81.0 Reading Disorder (dyslexia).	Impairment in reading achievement (accuracy, speed or comprehension).	Reading: Associated with Maths and writing disorders.
F81.2 Mathematics Disorder.	Impairment in mathematical calculation or reasoning.	Maths: A number of different skills may be impaired including "linguistic" "perceptual" and "attention" skills. Associated with reading and writing D.
F81.8 Disorder of Written Expression.	Impairment in writing skills	Writing: ". some evidence that language and perceptual-motor deficits may accompany this disorder".
80.1 Expressive Language Disorder. F80.2 Mixed Receptive-Expressive Language Disorder.	"Impairment of expressive language development" "Impairment of both receptive and expressive and language development"	Associations: delay in reaching some motor milestones, Developmental Coordination Disorder; social withdrawal, Attention-Deficit/hyperactivity D. EEG abnormalities; dysarthric or apraxic behaviours; neurological signs.
F84.5 Asperger's Disorder	"severe and sustained impairment in social interaction" "...impairment in the use of body postures and gestures ...repetitive motor mannerisms (e.g., hand or finger flapping or twisting, or complex whole body movements)".	Various nonspecific neurological symptoms or signs may be noted. Motor milestones may be delayed and motor clumsiness is often observed". "..sometimes observed in association with general medical conditions.." (e.g., the following are listed for Autism: encephalitis; phenylketonuria; tuberous sclerosis; fragile X syndrome; anoxia during birth; maternal rubella

...

Table 3.2 *Examples of DSM-IV (2000) Disorders: Main and Associated Features (cont.)*

Code. Disorder	Essential or Main Feature	Associated Features & Disorders
F90.0 Attention-Deficit/ Hyperactivity (combined) F98.8 Inattentive	Inattention; hyperactivity; impulsivity "fidgets" "squirms" "on the go" as if "driven by a motor"	"Impulsivity may lead to accidents (e.g., knocking over objects, banging into people). Oppositional Defiant D. Conduct D. Mood D. Anxiety D. Learning D. Communication D. Tourette's D.. Exposure to neurotoxin, infections or drug exposure in utero. Low birth weight
F95 Tic disorders Tourette's Disorder	"Multiple motor and one or more vocal tics"	Obsessive-Compulsive D; Attention-Deficit/Hyperactivity and Learning Disorders. Tics may interfere with daily activities (e.g., reading or writing).
Obsessive Compulsive D.	Recurrent and persistent thoughts Repetitive behaviours	Interfere with normal routine, occupational or academic functioning
F 91 Conduct Disorder	Aggression against people or animals; Property destruction; Lying or theft; serious rule violation	Attention-Deficit/Hyperactivity D. common. Learning D. Communication D. Anxiety D. Accidents rates appear to be higher
F91.3 Oppositional Defiant Disorder	"Recurrent pattern of negativistic, defiant, disobedient hostile behaviour".	High motor activity. Attention-Deficit/Hyperactivity Disorder. Learning Disorders and Communication Disorders.
M35.7 Familial Ligamentous Laxity	Hypermobility Syndrome.	Excludes: Q79.6 Ehlers-Danlos Syndrome

To the relatively inexperienced professional, the idea that one could easily use a manual like DSM to put children into neat categories might be quite appealing. Further experience, however, would reveal that such an exercise is not so easy. There have been two responses to the efforts of the APA and WHO in this regard. On the one hand we have Kaplan and colleague's approach (Kaplan et al., 1998; Kaplan 2005) which rejects the idea of continuing to try to separate one syndrome from the next. Instead, they suggest that a better way forward is to lump all disorders such as DCD, SLI, AS, dyslexia etc., into one bag with a purported common aetiology which they call atypical brain development (ABD). Although Kaplan disputes the idea that her position takes us back full circle to the long discredited notion of (minimal) brain damage/dysfunction syndromes proposed by Strauss and colleagues in the 1940s, the difference is contentious. That said, no one really doubts that for these children

something atypical has occurred and/or is continuing to occur and alter their developmental course. In their discussion of comorbidity reviewed earlier, Angold et al. (1999) also consider the problems inherent in the current APA and WHO diagnostic systems. In contrast to Kaplan, however, they stress the need for continued effort to distinguish quantitative and qualitative aspects of individual symptoms in relation to separate diagnoses.

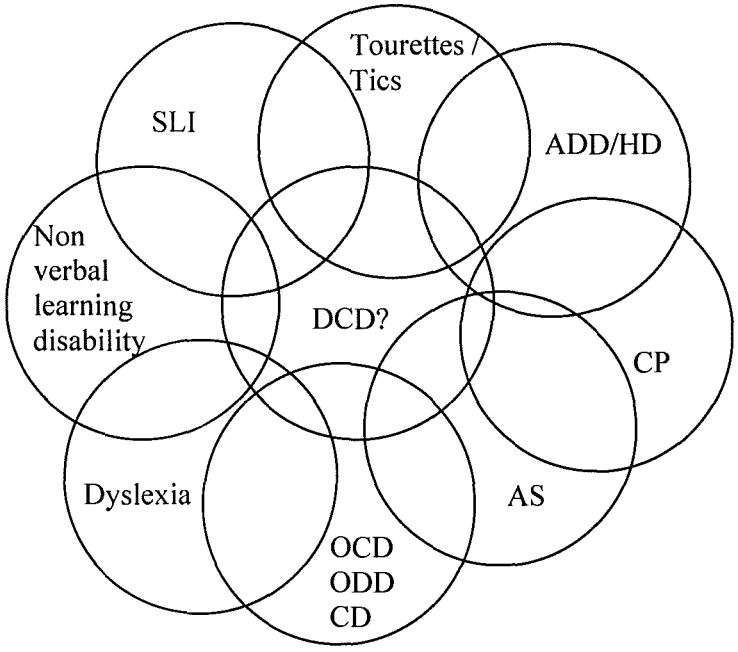
With regard to DCD, the condition at the core of the present thesis, there is no doubt that more quantitative and qualitative knowledge of 'clumsiness' in other developmental conditions would assist in the debate about whether DCD exists at all as a separate syndrome or is really just a symptom of many different developmental diseases and disorders. For instance, if one knew that clumsiness experienced by children with a speech and language problem or those with poor social interaction was very different from that experienced by children with 'pure' DCD, one would perhaps plan intervention for the two groups quite differently. At first sight, the literature on this topic might seem immense. For every single childhood condition mentioned so far, there are numerous papers which confirm that 'clumsiness' is a common feature. On closer inspection, however, one finds that few studies actually describe the movement problems in any detail and even fewer compare the chosen target group with a DCD group. Such a comparison is the starting point for the final study in this thesis.

3.5 The literature on co-morbidity, developmental disorders and DCD

The literature on comorbidity is truly vast and as Angold (op cit.) commented has grown exponentially over the last decade. From this literature, three key points emerge quite clearly: (i) there is overwhelming evidence that most conditions do co-occur, (ii) that the overlap between conditions can exceed 50%, and (iii) generally when more than two conditions co-occur/overlap there is a compounding effect on function (Kaplan et al., 1998; Gillberg, 2003; Visser, 2003). Providing more than a general estimation of overlap between conditions, however, is fraught with problems – use of labels, sample source, inclusion/exclusion criteria and measures used, to name just a few. In addition figures depend upon which way one examines the co-occurrence DCD with ADHD or ADHD with DCD! It is not possible in two dimensions to represent the complex relationships between overlapping childhood

disorders adequately but Fig 3.1 gives a rough idea of the extensive overlap one might find with DCD and various other common childhood conditions.

Figure 3.1 *Overlap of DCD and various childhood conditions*



Space does not allow an exploration of every possible combination of developmental disorder in which 'clumsiness' of movement is mentioned. In order to illustrate the way the co-occurrence of **movement** and other difficulties have been treated in the research literature, therefore, DCD is examined in relation to two commonly co-occurring disorders - ADHD and dyslexia. Other combinations were rejected, either because they are dealt with later in Chapter 9, or because the relevant literature is rather sparse.

(i) DCD, ADHD plus...

When one takes just Attention-Deficit/Hyperactivity Disorder (ADHD) and it's reported overlap with other conditions one is immediately faced with a diverse literature, which envelops both childhood and adult disorders. Not only the plethora of publications related to MBD (in effect the forerunner of the term ADHD) (Gillberg, 2003) but also more recent literature on substance misuse, conduct disorders (CD), obsessive compulsive (OCD) disorders, learning disorders and

criminal offending becomes relevant. Reports of an association between ADHD and DCD (or related terms) are numerous (Gillberg et al., 1983; Gillberg & Gillberg, 1989; Rasmussen & Gillberg, 2000; Hellgren et al., 1993; Landgren et al., 1996; Kadesjo & Gilberg, 1998; 2001; Piek et al., 1999; Pitcher et al., 2003; Harvey & Reid, 1997; 2003; Kooistra et al., 2005) and this is recognised in both the APA and WHO manuals (see Table 3.2).

DSM-IV sub-divides ADHD into ADHD Combined type (F90.0), ADHD Predominantly Inattentive Type (F98.8) and ADHD Predominantly Hyperactive-Impulsive Type (F90). Several studies have explored the functional profiles of children with ADHD and the results support sub-types within the broader picture. ADD combined with hyperactivity seems generally to link with a more pervasive condition than either inattentive or impulsive sub-types. There is a high prevalence of tic disorders in children with ADHD and the more severe Tourette disorder is not uncommon (Gadow et al., 2002). Again, ADHD plus tics or Tourette's reflects greater dysfunction than either tics or ADHD alone. In turn, tic disorders and ADHD are associated with obsessive compulsive disorder (OCD) and there are reports of co-occurrence of conduct disorder (CD) including oppositional defiant disorder (ODD). As noted above, it seems clear that when there are two or more comorbidities, both everyday life functioning and prognosis are more serious. Although attention is drawn to the association between ADHD and DCD, the DSM-IV manual proposes that the motor difficulties are the result of distractibility and impulsivity rather than impairment of motor function per se. Whether this is actually true or not has not been empirically established and was one of the questions that I had hoped to address in the final study (see below).

One of the most important investigations of the relationship between DCD and ADHD is undoubtedly, the longitudinal series by Gillberg and colleagues in Gothenberg, Sweden (see Gillberg, 2003). Kadesjo and Gillberg, (2001) report 87% of children meeting full criteria for ADHD had one or more, and 67% at least two, comorbid diagnoses. The Gillberg studies found that half of children with ADHD also met criteria for DCD and conversely 50% of those with DCD were identified with ADHD. To reflect the overlap between disorders including, DCD and ADHD, Gillberg coined the umbrella term DAMP (Disorders of Attention Motor Control and Perception), which remains in use in Sweden today. Sixty-six percent of severe cases

of 'DAMP' were identified with an autistic spectrum disorder and there appeared to be greater risk in the combined 'DAMP' condition than if either ADHD or DCD presented in isolation. In a more recent paper (Gillberg 2003, p. 908) Gillberg comments however, that: "DCD remains the 'black sheep' in the history of ADHD. It is perhaps the most common (and possibly the most specific) overlapping condition in ADHD; yet it is usually not even mentioned in assessment and intervention manuals for ADHD". He cites a Scandinavian study by Adler (1982) who reported that 'children with DAMP comprise the largest subgroup of all those who abstain/refuse/do not participate in physical education'.

As another example of a study, which focussed first on children with non-motor problems and identified some 'pure' case of DCD it is useful to look more closely at the Kaplan study (Kaplan et al., 1998). An initial cohort of 224 children, recruited on the basis of learning and/or attentional problems (but **not** specifically referred with motor skills concern) and a comparison group of 155 children were tested blind on robust standardised psycho-educational and motor tests. Strict criteria for ADHD, RD (dyslexia) and DCD were applied. One hundred and sixty two children had complete data sets but 47 children were excluded as they met none of the three conditions. Diagnostic categories and comorbidity was examined in the final sub-group of 115 children with complete data who met criteria for at least one of the attention, reading or motor conditions. Results showed that there was a total of 81 children (70%) with DCD compared to 48 (42%) with ADHD and 71 (62%) with RD. Results further revealed that fifty-three children (46%) had a single, 'pure' diagnosis compared to 54% with two or more conditions (34% with two disorders and 20% all three conditions). Interestingly from the initial index referrals 61 (27%) met criteria for DCD but also 20 (13%) from matched control groups with **no** apparent attention or learning problems were also identified with DCD. Whether the children derived from the controls formed the majority of the 'pure' DCD cases (26 in total) is not clarified.

In summary, both the Gillberg and Kaplan studies approached the issue of comorbidity primarily from the AD/HD 'plus' rather than the DCD 'plus' angle. It is much harder to find any research that focuses attention specifically toward children with pure DCD and compares them with a cohort identified with ADHD. This may be because DSM-IV criteria for AD/HD are more clearly defined and generally

easier to apply than the criteria for DCD. Another reason is that professionals who look at motor development often stem from professions such as physical education, human movement science, physiotherapy and occupational therapy where there is less funding and research tradition. One aim of Study 5 in the present thesis was to clearly identify groups of children including a group who have 'pure' DCD and another with ADHD.

When DCD is identified concurrently with ADHD and disorders associated with it, an important question arises. Is the movement difficulty a feature of and in effect the 'baggage' attached to ADHD behaviour or is the problem truly one of motor execution and/or planning? For example a hyperactive child may be unable to balance steadily on one leg as wriggling and hopping behaviours predominate over the calm still behaviour requested. The child has difficulty inhibiting his or her level of arousal. Another example is a child who may succeed on a motor task such as writing, provided there are no competing demands on attention such as a request to write quickly or to compose a story rather than copy a set piece. Similarly, other parallel cognitive demands can influence a child's motor performance, which may be compromised due to his or her preferred strategy which differ from the rules dictated by the assessment manual. This may lead to an odd, idiosyncratic performance but not necessarily lacking in motor coordination. Unfortunately, studies which are specifically designed to investigate questions relating to the way different facets of children's difficulties interact are sadly lacking.

The second example of how comorbidity as it relates to DCD has been studied in children turns away from ADHD to a different condition, also commonly reported in children with DCD, Dyslexia. The main feature of Dyslexia or reading disorder is listed in DSM-IV as "Marked impairment in reading achievement (accuracy, speed or comprehension)" and it may be associated with maths and writing disorders, the latter having links to DCD (accompanying motor-perceptual deficits). Furthermore disorder of written expression may be associated with language deficits (phonological disorder, expressive language disorder and mixed receptive-expressive language disorder) (see Table 3.2).

(ii) DCD, SLI, Dyslexia, and Dysgraphia

The co-occurrence of reading and writing difficulties, speech and language and movement difficulties was mentioned long ago by Orton (1937). Now in 2006, examination of DSM-IV reveals that DCD is listed as **associated** with SLI which itself features in writing disorder, which is in turn linked with reading. Empirical evidence for increased prevalence of DCD in children with specific language impairment (SLI) and high percentages (around 60%) of children with DCD who also demonstrate SLI are consistently reported (Cermak et al., 1986; Powell & Bishop, 1992; Hill, 1998). Indeed, SLI might well have been chosen for further examination in this thesis as clinically, one of the first concerns raised by parents that often heralds DCD is mild language delay or articulation difficulty. Many of the children diagnosed with DCD at school age have already met with a speech and language therapist in infancy, who is frequently the first professional to have assessed the child. In a small physiotherapy intervention study we found that over half of our participants had a history of a speech and language problem (Peters & Wright, 1999). Similarly, Stephenson et al. (1990) reported that 52% of parents of a group of children aged between five and eleven years, with motor/learning difficulty referred to occupational therapy had had speech difficulties usually recognised before school entry. Once again though, overlap is not 100% and it is clear that not all children with SLI or dyslexia have DCD and vice versa not every child with a developmental motor coordination problem will present with SLI or reading difficulty. However, both reading and writing difficulty are reported frequently in children with DCD (e.g., Henderson & Hall, 1982) and vice versa in children labelled dyslexic (Geuze & Kalverboer, 1994; Ramus et al., 2003). Dysgraphia or writing difficulty is prevalent in children with SLI, DCD, ADHD, RD AS and other developmental disorders (O'Hare & Brown, 1989; Barnett & Henderson, 2005). Writing difficulty was reported in 40% of a group of children attending a rheumatology clinic for hypermobility (parent report) (Adib et al., 2005) although it is not clear whether the difficulty was related to associated joint pain or motor impairment.

In contrast to the literature on ADHD, which seems disproportionately confined to reporting prevalence figures, there have been one or two studies which have set out to look at the motor difficulties in children with SLI, Dyslexia and/or DCD in a different way (Powell & Bishop, 1992; Hill et al., 1998; O'Hare & Khalid, 2002; Ramus et al., 2003). For example, Hill et al., 1998, hypothesised that errors in

gesture production displayed by children with DCD and with SLI might reflect differential features of underlying planning or praxic dysfunction. Their study examined three groups of Primary school children: A group of children with SLI, a group with DCD, an age matched comparison children (mean age 9-10 years) and a further control group of younger children aged 5-6 years. What they found was that the gestures of children with both SLI and DCD resembled those of younger children especially when performed to verbal command and although children with DCD tended to be poorer on imitative gesture, qualitative differences in error type were not identified. The authors conclude that the results suggest immaturity in praxis development that is common to SLI and DCD rather than specific to either group.

A rather different approach to studying the links between dyslexia and DCD can be found in the recent work of Nicholson and Fawcett. Whereas other researchers have approached dyslexia from the perspective that language (especially phonological processing) and learning to read are closely associated and therefore depend upon similar underlying mechanisms, Nicholson and Fawcett (1990), Fawcett and Nicholson (1992; 1995; 1999), Fawcett et al. (1996) take a different view. Their automaticity/cerebellar hypothesis proposes that motor control and therefore speech articulation are sub-served by the cerebellum, and as the cerebellum is important for automization of learned skills (such as reading and writing), dysfunction at the cerebellar level will disrupt phonological processing. Thus, they suggest that a fundamental link between movement and reading impairment may be mediated by the cerebellum, and therefore other traditional signs of cerebellar dysfunction such as truncal ataxia, balance problems, dysmetria may be not unexpected. To test their hypothesis, they undertook a series of studies comparing children with dyslexia and matched control groups on tasks which were chosen as reflecting cerebellar function – time estimation (Nicholson et al., 1995), motor skill (Fawcett & Nicolson, 1995) and a combination of tasks they described as cerebellar and cognitive (Fawcett & Nicolson, 1999). They included perturbation of static balance with vision or with vision occluded, arm shake to determine motor 'floppiness', toe tapping speed along with phonemic segmentation, nonsense word repetition and picture-naming speed. They concluded that 95% of the dyslexic participants were 'at risk' on one or more 'cerebellar' item. There are many criticisms one could make of this work, not the least of which is the attribution of many of the processes measured to cerebellar function. For example, almost all (51/59) of the dyslexic children demonstrated low

muscle tone as measured on the very subjective 'arm shake' whereas this was apparent for only 8 out of 67 of the control group. A problem with the measure of muscle tone used in this study, however, is that failure might equally reflect body composition or flexibility, which in itself might compromise balance especially in a child who avoids physical activity and is generally less fit. In an empirical test of the Fawcett et al. hypothesis, Ramus et al. (2003) used a multiple case study to assess leading theories of developmental dyslexia in 16 university students with dyslexia and 16 controls matched for age, gender and IQ. They found little support for the notion that motor impairments, when found, have a cerebellar origin or reflect an automaticity defect. However what is of interest in the Fawcett studies as far as this thesis is concerned is the methodology used. Their technique of examining static balance using a dual motor-cognitive task might be a useful way of studying how 'clumsy' children with different developmental conditions cope with the differing demand of tasks like these.

3.6 Summary and Conclusion

There is undoubted evidence of common symptoms across many of the developmental disorders. One of the questions one needs to ask next is how these common symptoms can be accounted for theoretically and dealt with practically. The concepts of DAMP and of ABD suggest common underlying links which may be at a very basic level. Kaplan's concept of ABD, in particular, although seemingly a retrograde step to MBD, does draw attention to advances in understanding of brain development, neuro-anatomy, physiology and psychology at a cellular and genetic level, with particular reference to the dynamic interactions between macro and micro systems and environments. To pursue this line of enquiry further, however, requires longitudinal studies, sophisticated measurement tools and imaging techniques, and might also utilise recent information emanating from genetic studies. Attractive as it might seem, this is far beyond the scope of this thesis.

Earlier in this chapter, the parents and professionals perspective of co-existing conditions was discussed. This focussed on co-occurring symptoms reported by the parents and signs common to several syndromes, observed by professionals. Here the features of the disorders were dealt with at a purely functional or phenotype level but they did refer to complex and dynamic interactions between them. A problem here, however, is that interactions are mostly dealt with at a descriptive level and

very few studies look at interactions between the symptoms characterising various developmental conditions directly e.g., attention on movement, movement on social interaction. One of the aims of the final study in this thesis is to examine such interactions in more detail.

Chapter 4
Aims of the Research

4.0 Introduction

The previous three chapters provided an overview and critical analysis of the literature upon which the present concept and understanding of DCD is based. The breadth of the literature relating to 'clumsiness' in children demonstrates that the subject is not neatly ring-fenced into medicine, education or psychology but spans all of these plus many other fields. One of the problems that emerges throughout the discussion, therefore, is how one deals with a subject which spans different fields when the fields themselves have historically remained largely isolated one from another? In the past, the way 'clumsiness' was conceptualised, investigated and treated developed along different paths in health and education. Over the last decade, however, the gulf between them has, in most countries, gradually narrowed, and communication across the divide has led to sharing of the many problems, which still need to be solved in this area. The studies included in this thesis were conceived in the context of these shared problems and the purpose of the present chapter is to outline the main aims of the five studies which follow. The rationale and general aims of each study are presented, followed by a brief discussion of the methods adopted. More specific objectives are developed in the introduction to each study.

4.1 Study 1: Three common terms used to describe children with mild to moderate movement difficulties - how are they defined and perceived by health and educational professionals in the UK?

Chapter 1 outlined the background history, derivation and developmental path of the various terms used to describe 'intelligent children, whose motor development is unexpectedly slow'. In the course of describing the labels used over the years, this review highlighted some of the problems that arise when the terminology employed extends from common English terms through specific professional 'languages' to the language of official classification systems. It also showed that variation in the degree of knowledge, understanding and application of labels continues today, both within and across health and educational circles. Until these issues are resolved, standard protocols for identification and intervention by professionals working as multi-

disciplinary teams cannot be developed. In addition, progress in research is undermined when it is unclear whether studies purportedly on the same topic are actually comparable.

Although recent reviews suggest that professionals may use diagnostic labels either interchangeably and/or according to their professional discipline (Missiuna & Polatajko, 1995), there has been little formal analysis to support the observations. Put another way, although many people have listed the numerous labels used to describe children with specific difficulty in the motor domain there have been few objective attempts to compare the perception of such terms by professionals from different fields. The aim of Study 1 (Chapter 5) therefore, was to examine how UK professionals presently use, understand and interpret various terms or labels for children with idiopathic movement difficulties. Specifically, the study attempted to determine objectively the perception of three terms by doctors, therapists and teachers currently practising in the UK. As part of this exercise, questions were asked about the familiarity and acceptability of labels such as DCD and an attempt was made to identify any trends in the agreement or disagreements identified across the perception of terms.

4.2 Study 2: Labels and service provision - an investigation of parent's perceptions

In the UK, the roots of DCD can be traced back to the identification of a 'clumsy child' syndrome which led eventually to the recognition of a need for service provision for the families concerned. Longitudinal studies have shown that very few, if any, 'clumsy' children simply grow out of their problems and that their difficulties often persist into adulthood. Central to the management and delivery of intervention for children with DCD, therefore, is what exactly is important to the families themselves and how they perceive issues such as labels, diagnoses and access to intervention. As Study 1 will show, there continues to be confusion in the application of labels and this can have implications for access to intervention.

Clinical audit is a part of evidence-based practice in physiotherapy and thus the aim of the second study was to harness and extend a procedure already in place to examine parent/carer satisfaction with the service provided in the centre where the

current researcher worked. Although the subject of diagnostic labels has spawned numerous publications and stimulated heated discussion at conferences, very few researchers have considered the issue from the parent's perspective. Consequently, one aim of Study 2 was to extend the investigation of labels from the professional's viewpoint addressed in Study 1 to encompass the parent's perspective.

Reports have indicated that not only is there no clear 'best' intervention for children with DCD but also the route through referral, assessment and intervention is not equitable. The experience and views of parents are thus important contributions to improving this situation. Study 2 aimed to examine qualitatively the perceptions and experiences of real families by gathering and analysing their comments on their journey through referral, assessment and intervention offered by one service provider.

4.3 Study 3: The difficulty of diagnosing and describing DCD: A retrospective study

One of the clearest messages to emerge from the literature review was that DCD is difficult to define - and therefore, to diagnose. In addition, it was noted that the official classification systems in existence do not help much with this problem. The DSM-IV and ICD-10 manuals list criteria but give no precise indication as to how these should be applied. This problem is not unique to DCD, but is common to all developmental disorders especially the group, often labelled 'Specific' learning difficulties (Whitmore & Bax, 1999). In most instances, there is on-going discussion to try to clarify and standardise the criteria and their interpretation.

Being based at a specialist paediatric hospital, the present researcher was in an ideal position to evaluate one way of operationalising the criteria for DCD proposed in DSM-IV. The physiotherapy department collected both standardised and non-standardised data in a way that made derivation of an algorithm fairly straightforward. By applying this retrospectively to a large cohort of children, referred to GOSH because of concern regarding their coordination it was possible to examine the outcome in relation to current thinking about DCD as a separable syndrome. For most researchers, DSM-IV Criterion C presents a particular problem as they do not have access to the expertise necessary to apply it. As noted earlier, the medical literature shows that very many childhood conditions feature movement

difficulty and more specifically 'clumsy' symptoms. There is often a real problem in defining clear boundaries between normal and abnormal development in the light of normal variability, and between and within syndromes that have symptoms in common. This study offered the opportunity to determine how many children would meet DSM criteria if Criterion C were strictly applied and to discuss implications of this for future research.

The literature suggests that most clinicians and researchers believe that sub-types exist within DCD although to date confirmatory evidence has been insufficient. The present study included a large enough sample of children with complete data sets to examine this question further.

Unlike the previous two studies, this study took the form of a retrospective case-note review, which focussed on a group of children referred to a specialist paediatric hospital because of movement difficulty or putative DCD. Data available included results of two standardised perceptual-motor tests and a handwriting assessment, data on age, socio economic status (SES), IQ and birth history, as well as detailed clinical observations. Specifically, the aims of the study were (i) to evaluate the DSM-IV entry for DCD by examining the effectiveness of one interpretation of the four diagnostic criteria proposed, and (ii) to reconsider the question of subtypes within the syndrome of DCD.

4.4 Study 4: The value of the single case study: What real life examples can add to our knowledge of DCD

The 'clumsy child syndrome' or DCD is recognised by its entry in DSM-IV but no two children present in an identical manner. Single case studies provide an opportunity to highlight specific features and details that may be lost in group data but may be of considerable importance if we are to further understand a condition and develop effective intervention.

Studies reviewed earlier alluded to individual differences in the problems of children with DCD but the present study allowed more detailed examination of profiles and trajectories of particular children. These personal histories address specific questions in a new way and are designed to illustrate theoretical and practical issues raised in the thesis. The aim of the case studies presented in Chapter 8, therefore, was to put

the problems that researchers have raised into the wider context by including comment not only on the motor difficulties the children experienced but also to introduce data on the other commonly occurring problems these children encounter.

4.5 Study 5: DCD: A separate syndrome or not?

Studies 1-3 of this thesis proceeded on the assumption that DCD existed as a separable syndrome and support for this idea emerged from Study 3. However, one limitation to this study was that information from other domains of behaviour was not available to the researcher as data were collected retrospectively. Thus, it was not possible to determine whether children who met all four criteria for DCD might also have met criteria for other conditions such as Attention Deficit Hyperactivity Disorder (ADHD) or Asperger's Syndrome (AS). In Chapter 3, the literature review highlighted the fact that in developmental medicine and neuropsychiatry the existence of children with only one 'major' symptom is rare. Far more common are children who have a whole range of symptoms which vary in their severity and dominance over time. The review then pointed to longstanding theoretical and practical problems that this overlap has caused. Having touched upon the fact that children who meet criteria for DCD experience difficulties in other domains of behaviour, this chapter focussed more closely on the overlap or association between DCD and other conditions.

In Chapter 3, the idea that DCD might be better conceptualised as one phenotypic presentation of a common core brain abnormality at a genetic or biochemical level was introduced. It was not possible in this thesis to explore the genetic or biochemical origins of these conditions. However it was possible to look more closely at the movement profiles of children who carry different labels, as a means of exploring the similarities and differences between them. There have been a few comparisons of groups of children bearing different labels. However, these studies nearly always compare only two groups and some of these are not very well defined. Consequently, the aim of this study was to examine clearly defined groups of children each with a different diagnosis, to see if they may be uniquely identifiable by their movement profile.

In order to achieve this objective it was essential to begin by looking in further detail at differential diagnostic issues. Although children may be referred to a particular

clinic/expert because of concern for one specific aspect of their behaviour, it cannot simply be assumed that they will eventually carry the label linked to that centre's expertise. In this final study, therefore, one objective was to examine the 'fit' between referral source and diagnostic category. Once a group of children with relatively clear diagnoses had been identified, two approaches were taken to examining similarities and differences in their movement profiles. The first was entirely descriptive. Standardised measures were plotted on graphs and 'eyeball' comparisons made. The second, and more robust approach was to test specific hypotheses about possible differences between the groups using the techniques of experimental psychology. For instance, attention has recently been drawn to the overlap between children with Benign Joint Hypermobility Syndrome and DCD. Since such overlap is not likely to be 100%, one might specifically test whether hypermobility adds to the effect of poor coordination, by itself, and if so, on what tasks. For instance, one might hypothesise that hypermobility would affect balance but not manual dexterity. In this final study, a series of questions are asked, taking different subsets of children from the larger group of those with movement problems. A comparison group of typically developing children also took part.

4.6 Methodology used in the research studies

The choice of methodology for any research study is mainly dependent on the type of question being asked. Broadly speaking, research designs fall into two categories, group/ cohort designs and single case studies, each of which can be equally robust in its own way. Put simply, the more commonly employed group study provides information about features common to that specific cohort as a whole and individual characteristics are lost. In fact, any individual who seems markedly different from the rest of the group is likely to be excluded, on a statistical basis, as an outlier. In contrast, the single case study takes individual variation as its starting point and provides information about a particular individual, which may be unique and possibly not generalisable to the population as a whole. Since the information provided by different kinds of study is complementary rather than exclusive, many large-scale research projects combine both methods in a triangulation approach to test a particular hypothesis in different ways and help verify the data obtained. Whatever the methodology selected all must rely upon strict adherence to carefully standardised protocols in order to obtain valid, reliable data.

The studies designed and reported in the present thesis addressed a series of questions that required different methodologies. Since the first two studies were concerned with opinions a questionnaire design was deemed appropriate. Questionnaire techniques generally use interview, telephone and postal approaches for data collection. Questionnaires may be delivered face to face, at a distance (telephone or postal) and as a group or individually. Questions may be closed or open ended and can differ on ordering and scaling depending on the aims of the measurement and method of analysis chosen. Study 1 used an open-ended self-completed questionnaire format presented to individual participants. The analysis of the results utilised content analysis to interpret the rich qualitative data and non-parametric statistics to test for significance. A questionnaire was also chosen for Study 2 but in this instance a postal method was considered more appropriate. The questionnaire was designed specifically to meet the needs of audit of the Physiotherapy Service and again was a group study but each questionnaire was completed individually. A mixture of questions requiring both forced choice (closed) or open-ended responses enabled a variety of data to be collected.

For Study 3, the design used a cohort cross-sectional method but it was convenient to employ already collected data retrospectively in order to access previously documented records of a large number of children. A wide range of quantitative and qualitative data analysis was examined. The technique of cluster analysis was introduced in order to consider the possibility that DCD might be better conceptualised as a syndrome with multiple subtypes.

As noted above, single case studies provide information that cannot be, or is difficult to, extract from group designs. Whereas all the group studies in the thesis were cross-sectional examining data collected at one point in time, the most important feature of Study 4 was the inclusion of longitudinal case studies where the child was followed up over a number of years.

Finally Study 5 was an extension of Study 3, in that some of the measures and analyses were identical. In most other ways, however, Study 5 was much more complex and unique in that it involved a number of design features and techniques, which had not previously been used in this area of research. Of particular importance, was the introduction of single blinding in order that the diagnosis of

participants was not revealed to the researcher prior to, or during any of the data collection, thus helping to reduce possible experimenter bias. Additionally, the assessment focused on wider domains of function and included several novel assessment items specifically designed for this study. These included both published standardised tools including parent-completed questionnaires, and also shorter questionnaires and perceptual motor tests designed especially for the study. In contrast to Study 3, which was retrospective, Study 5 recruited participants prospectively. It was a multi-centre trial and although more extensive ethical approval was therefore required, this allowed for participants to be recruited from a wider range of clinical and non-clinical sources.

Chapter 5

Study 1: Three common terms used to describe children with mild to moderate movement difficulties – how are they defined and perceived by health and educational professionals in the UK? [1]

5.0 Introduction

Many professionals find discussions about terminology tedious. "What does it matter what we call them, as long as we describe the problem clearly and can do something about it?" However, the choice of labels has important implications for both theory and practice in the fields of health and education (Warnock, 1978; Davies, 1994; Gardner-Medwin, 1995; Bax, 1999; Hart, 1999). At a theoretical level, confusion about terms and their definitions can frustrate scientific research by leading to inadequate and inconsistent criteria for defining samples and thence to difficulty in comparing one study with another. Accurate definitions underpin the collection of statistics for epidemiological studies, for planning intervention and for monitoring progress. At a practical level, we are all familiar with the arguments for and against giving children labels, which signify that they are in some way different from the 'norm'. On the positive side, a condition defined in a certain way may confer special entitlement to benefits and services. On the negative side, a label may be difficult to shed, even when the child has changed and the description no longer applies.

Clarity about terminology also matters in relation to the collection of statistics, which inform policymaking, nationally and internationally. The advent of Clinical Governance and evidenced-based practice in the UK (DFE, 1994; McKinley, 1996; NHS, 1999; CSP & COT, 1999) has highlighted the need for a nationally standardised system of classification. A collaborative project between the USA and UK has the even more ambitious objective of producing a "standardised healthcare language throughout the English speaking world" (DOH, 1999) (see Chapter 1). In

[1] A version of this Chapter has been published. Peters, J. M., Barnett, A. L., & Henderson, S. E. (2001). Clumsiness Dyspraxia and Developmental Coordination Disorder: How do health and educational professionals in the UK define the terms? *Child: Care Health and Development, 27,* 399-412.

the area of motor difficulties in children, however, we are a very long way from reaching a conclusion on the use of terms and their interpretation.
Recent reviews of the various labels applied to a discrete childhood syndrome which has 'clumsiness' of movement as its defining symptom have drawn attention to confusion in the present use of terms (Henderson & Barnett, 1998; Polatajko, 1999). For some professionals, different diagnostic labels seem to be used interchangeably. For others, the different labels are used to refer to slightly different conditions and may be coloured by the particular background of the user (Missiuna & Polatajko, 1995).

The study now reported in this chapter was designed to shed light on the current state of knowledge with regard to understanding, interpretation and current use of labelling in the UK. The aim of the present study therefore was to determine how UK professionals from health and education perceive various terms used in relation to children who lack the ability to move fluently. Three terms, 'clumsy' 'dyspraxia' and 'developmental coordination disorder' were chosen which were deemed to represent labels used since the notion of a separate syndrome was first proposed. The first of these terms, 'clumsy' was chosen on the grounds of historical precedence and common descriptive usage. The second was employed because of its roots in medicine and its adoption by the Dyspraxia Foundation, the parent lobby now active in the UK. For a term with 'official blessing', 'Developmental Coordination Disorder' (DCD) from DSM-IV (American Psychiatric Association, 1994; 2000) was chosen rather than 'Specific Developmental Disorder of Motor Function' (SDD-MF) from ICD 10 (World Health Organisation, 1992) as the former is much more commonly used by researchers around the world (Fox & Polatajko, 1994; Barnett et al., 1998).

In summary the aims of the study were: 1) to compare and contrast different professionals' familiarity with the three terms and the extent to which they found them acceptable, and 2) to characterise any systematic similarities and differences in the perceptions of each term. To achieve this end, definitions of the three terms were collected using a self-administered, free-response method, which offers participants freedom to express their ideas. Although analysis of such data produced by this technique is time-consuming, it has the advantage of capturing the full richness of the content (Glaser & Strauss, 1967; Oppenheim, 1992).

5.1 Method

5.1.1 Participants

The 234 professionals taking part in this study were a convenience sample from health (57%) and education (43%) attending meetings or courses across the UK in 1998. None of the meetings assumed any prior knowledge of the subject of interest in the present study.

5.1.2 Procedure

Response sheets were distributed to participants by the present researcher and three colleagues. Respondents then wrote down their own definition of each of the three terms, in the presence of the researcher, without consultation. Professional status was recorded and the form returned immediately.

5.1.3 Development of the coding frame

Table 5.1 shows typical examples of the responses obtained for each of the three definitions.

Table 5.1 *Two Examples of Individual Responses*

CLUMSY: "General term for a mildly uncoordinated child who often bumps into people, trips, drops things and has poor motor control." DYSPRAXIA: "Motor planning disorder of neurological origin in the absence of muscle weakness. May affect oral motor skills and therefore speech as well as fine and gross motor skills." DEVELOPMENTAL COORDINATION DISORDER: "No idea!"
CLUMSY: "Mild coordination problem, "cack-handed" DYSPRAXIA: "Difficulty in planning, organising and carrying out activities. Includes coordination, sequencing and perceptual difficulties" DEVELOPMENTAL COORDINATION DISORDER: "As dyspraxia"

In order to capture the variation in the content of the 702 definitions provided, it was necessary to develop a reliable coding frame. This was done by the technique of content analysis recommended in Oppenheim (1992), which involves the sequential checking and re-checking of a category system until coding agreements are maximised and disagreements minimised.

Professionals from four different disciplines, occupational therapy, physiotherapy, psychology and education took part in this process. Initially 40 responses were randomly selected and examined by the present researcher in order to establish familiarity with the general content and become immersed in the data. Several readings of the responses were made. Recurrent ideas/words/synonyms were grouped together. Themes were identified from the grouped words/phrases. These were given a primary classification heading. For example a theme of 'accidents' was drawn from recurrent words/phrases such as 'trips', 'knocks things over', 'drops things' 'mistakes'.

To determine reliability of these primary classifications the researcher coded a further random sample of 15 responses. A second rater (an occupational therapist) was asked to classify an unmarked response set using the same coding frame. Inter rater reliability showed 70% agreement. Responses that were difficult to classify by either rater were discussed. Adjustment was made to the coding frame. Further inter-rater reliability assessed on three occasions between 1-4 raters always using a random sample of questionnaires yielded 85% agreement. The final version of the 26 category coding frame is shown in Table 5.2. Intra-rater reliability of the final coding frame was also tested. The present researcher re-coded a random sample of 10 questionnaires following a two-week interval and achieved 85% agreement with her previous coding.

Table 5.2 *The Coding Frame: Examples of what was Included in Each of the 26 Categories*

CODING FRAME

FAMILIARITY and ACCEPTABILITY OF TERMS

1. Unknown term: e.g., "don't know", ?, no response
2. Synonym: e.g., indicates two or more of the terms as interchangeable
3. General term: e.g., a lay, or non specific term
4. Specific term: e.g., indicates a term is specific to a profession
5. Official classification: e.g., entry in ICD or DSM-IV
6. Medical condition: e.g., mentions "abnormality", or "disease" in definition
7. Developmental: e.g., refers to "delay", "immaturity" but not abnormality
8. Avoided: e.g., uses phrases such as "derogatory", "not used"
9. Mild: e.g., refers to condition as "not serious", "slight"
10. Severe: e.g., includes phrases such as "more serious", "wide effects", "complex"

"CORE" MOTOR ELEMENTS

11. Gross movement difficulty: e.g., difficulty with gross motor, general incoordination
12. Fine motor difficulty: e.g., specifies problems with manipulation etc.
13. Activities of daily living (ADL): e.g., mentions difficulty with dressing etc.
14. Accidents: e.g., uses terms like "trips up", "bumps into things", "drops things"
15. Motor control/execution: e.g., refers to force, timing, smoothness of movements
16. Motor planning: e.g., refers to "motor organisation", "praxis" etc.
17. Perceptual/sensory: e.g., visual/perception/proprioceptive/tactile, sensory integration
18. Spatial: e.g., uses phrases such as "misjudges distances" etc
19. Communication: e.g., refers to speech, language, articulation, oral/mouth
20. Lack of strength: e.g., mentions words such as "weakness", "hypotonia", "fatigue"
21. Laterality: e.g., refers to dominance or handedness
22. Brain/body link: e.g., uses words like "brain-action", "mind-body"

ASSOCIATED PROBLEMS

23. Cognitive: e.g., mentions phrases related to I.Q. memory, understanding
24. Behaviour: e.g., mentions problems with concentration, attention or conduct
25. Emotional/social: e.g., uses words such as "anxious", "nervous", "low self esteem"
26. Academic: e.g., refers specifically to "writing", "spelling", "mathematics" etc.

5.2 Results

Of the 234 professionals in the sample, 133 were from health, mostly paediatric specialists. The proportion of doctors was 32%, occupational therapists (OTs) 38%, physiotherapists (PTs) 22% and speech and language therapists (SALTs) 9%. Medical doctors included senior and junior grades. The majority of doctors were hospital based and most were paediatric trained. Consultant grades represented several different specialities: four paediatricians, paediatric neurologists, a neurosurgeon, a neuropsychiatrist, a geneticist, and a paediatric clinical biochemist. Therapists were predominantly paediatric specialists. The 101 educational professionals ranged from a retired head teacher, a lecturer, three special educational needs coordinators (SENCO), secondary and primary teachers in special and mainstream education.

5.2.1 Familiarity and acceptability of the terms

Respondents varied in the extent to which they were familiar with the terms. Whereas no one failed to provide some sort of definition of the term 'clumsy', 7% of respondents were unfamiliar with 'dyspraxia' and 32% with 'Developmental Coordination Disorder' (DCD).

All therapists, regardless of profession, were familiar with the term 'dyspraxia'. Of the 17 respondents who failed to offer a definition for the term all but two were teachers. A further twelve teachers also confused 'dyspraxia' and 'dyslexia': *"Similar to dyslexia but I can't remember the difference", " a physical version of dyslexia".*

With regard to 'DCD', 74 professionals were unable to offer a definition. OTs stood out as being most knowledgeable (only two failed to provide a definition). Nine participants, all therapists, knew that 'DCD' was the term selected by the American Psychiatric Association for its official publication. Lack of knowledge of the term was similar for teachers, doctors and PTs (37%, 36% and 31% respectively).

The term 'clumsy' was castigated as "*rather an old-fashioned term, largely replaced by dyspraxia*" and as "*a lay term*". Seventeen respondents avoided its use "*Not a term I use – prefer to use motor learning difficulties*", "*The term 'clumsy' .... should*

not be used as a clinical term these days". Ten others were opposed to the term 'DCD' e.g., "...I personally use dyspraxic term"

Lack of precision was referred to in a number of entries. For example, the term 'clumsy' was perceived as encompassing movement within the normal range e.g., *"Just a bit awkward ... can be a normal person!"* Similarly, an OT considered the term 'dyspraxia' to be used imprecisely: *"This term is often used by health professionals to describe general motor learning difficulties whereas it should be used in a more discriminative way i.e., dyspraxia on verbal command etc"*

5.2.2 Definitions, similarities and differences

Sixty percent of respondents provided a definition for each term. Of these, 11% indicated that all three were synonymous. Another 11% indicated that two were synonymous, usually 'DCD' and 'dyspraxia' (7%).

Some of the perceived differences between the terms seemed to be the product of key professional constructs. Of 'dyspraxia', for example, a doctor wrote *"Difficulty with fine motor control in the absence of paresis of the muscles involved in execution of the relevant movement."* whereas a PT wrote *"Co-ordination disorder often linked with proximal hypotonia"*, and an OT wrote *"a diagnosis within sensory-integrative disorder where the tactile and proprioceptive systems are underactive"*. Similarly, the teachers' concern with the classroom could be seen in the definitions *"Difficulty forming letters"* or *"Mixing letters around the wrong way in words"*.

5.2.3 The 'core' elements of the definitions

From the 234 participants, the total possible number of responses to the three terms was 702. There were 91 failures to provide a definition and these were discarded leaving a total of 611 valid definitions remaining. The content of these was then analysed with reference to the 12 categories in the Coding Scheme listed as representing the 'core' **motor** elements of the conditions (see Table 5.2).

The percentage of definitions that embraced fine motor difficulties scarcely differed across terms ('clumsy', 21%, 'dyspraxia', 18%, 'DCD', 21%). In contrast, for gross motor difficulties, the percentages were noticeably higher for the terms 'clumsy' (71%) and 'DCD' (64%) than for 'dyspraxia' (42%).

Handwriting was mentioned by 22 respondents (20 teachers, an OT and a PT but not by doctors). In 16 instances it was included in the definition for 'dyspraxia' and handwriting comments for 'clumsy' and 'DCD' were five and three respectively. The comments covered different aspects of handwriting: *"Awkward pen/pencil grip, uneven and inconsistent letter formations jerky hand movements instead of smooth flow"* ('clumsy'); *"...confusion in forming letters..."*(dyspraxia); *"...unable to form letters or imagine them"* (DCD).

There were surprisingly few mentions of difficulties with activities of daily living for any of the terms (< 10 in each case). For the 'accidents' category, the number of references to the term 'clumsy' (33%) was substantially higher than for either of the other two (4% in each case).

There was rather little variation in the number of references to control parameters, such as the force, timing, speed and accuracy of movement. The term 'clumsy' attracted 11%, 'dyspraxia', 22% and 'DCD', 14%. The phrases *"uncoordinated movement, not smooth and free flowing."* ('clumsy'); *"...difficulty making conscious controlled movements"* ('dyspraxia'); *"...poor coordination in term of poor motor control (not poor planning)"* ('DCD') illustrate the similarities between participants' views of each term. In contrast, the proportion of statements referring to motor planning was substantially greater for 'dyspraxia' (43%) than for the other two terms (6% and 11% respectively). Of the term dyspraxia, one respondent wrote: *"Unable to do what you want when you want, but then you can do it when you don't think about it."*

Sensory, perceptual or spatial difficulties were mentioned for all three terms. Sometimes, a single modality was specified e.g., *"Poor visuo-perceptual skills"*, "*tactile and proprioceptive systems are underactive.*" At other times, the integration of sensory and motor information was highlighted: *"Difficulty integrating sensory information"*. A number of respondents, mostly teachers, made statements which seemed to link perceptual and/or spatial problems to difficulties with handwriting.

The term 'dyspraxia' included 31 references to communication problems e.g., *"hears and understands language but cannot connect it to their own oral signals"* whereas fewer than four references were made for either 'clumsy' or 'DCD'. The three

remaining categories covering lack of strength etc, laterality, and brain-body links were referred to infrequently throughout, (10, 3 and 14 references respectively). Among the 14 references to a brain-body link one participant wrote: *"Where the connections between the brain + body don't work ... can do things from a motor point of view + a brain point of view but not together"; "An autistic type of disorder where body and mind fail to communicate".*

Severity also distinguished between the terms, being mentioned in connection with dyspraxia much more often than with clumsiness. – e.g., *"More pronounced than 'clumsy'.* Similarly, no one described 'clumsy' as a medical term, whereas 'dyspraxia' and 'DCD' were perceived as associated with a medical condition by around 10% of respondents. Of 'dyspraxia', one respondent wrote *"A pathological state, congenital or acquired, whereby the subject cannot access fully established motor routines...."*

5.2.4 Associated problems

All three terms attracted comments in all four categories listed under the heading 'associated problems' in the coding frame (see Table 5.2). Within the category, academic, 'dyspraxia' attracted 15% of responses compared to less that 6% for either 'clumsy' or 'DCD'. The percentages of statements did not vary appreciably across any of the other terms or categories (cognitive and emotional/social < 5% and behaviour < 10%). The nature of the comments made suggested that the term 'dyspraxia' was perceived as being broader and more complex than the other two. For example, 'dyspraxia' was described as: *"...associated with other specific developmental problems"; "A child who is 'clumsy' but also has other traits which affect learning issues"; "...linked usually with...spelling difficulties etc."*

References to cognitive functioning varied in both content and style. For example, the phrase *"piece of behaviour which could have been avoided with forethought"* (related to clumsy) seems to imply that the clumsiness of movement in itself might be caused by failure to plan at a cognitive level. This contrasted sharply with more global references to a discrepancy between cognitive and motor development as when one respondent referred to 'dyspraxia' as *"a significant discrepancy (> 20 points) on verbal/performance skills".* Another wrote of 'DCD' *"...acquisition of functional movement out of line with cognitive potential"*

Frequency of reference to behaviour problems appeared to be inversely related to their perceived severity. For instance, many of the words and phrases used to define the term 'clumsy' seemed to refer to a relatively minor degree of inattention or carelessness *" a bit scatty"*: *"...may be thought slapdash", "...due to inattention or carelessness". "..The class/family clown.."* In contrast, the term 'dyspraxia' attracted phrases like *"non-compliance", " behaviour difficulties", "disruptive", "anxious"* and *"poor self-esteem".*

Emotional/social aspects were mentioned infrequently for any term (< 5%). However, once again remarks relating to 'dyspraxia' or 'DCD' implied greater severity than those included under definitions of clumsiness. Of the few comments related to academic attainment or schoolwork, the majority occurred within definitions of 'dyspraxia'

5.3 Discussion

In the present study, the objective was to determine how professionals currently practising in the health and education services in the UK perceived three terms which have been used to refer to children with a 'specific' difficulty in the movement domain. Most participating professionals worked with children, but variation in discipline and experience was considerable. Health professionals included senior and junior grade doctors, and all grades of therapists working in clinical, research, acute hospital and community settings. Similarly participants from education included lecturer grade in academic institutions, a full range of teachers working in primary, secondary, special, mainstream and private education. Consequently, the results obtained should be generalisable to all health and education professionals currently working with children in the UK.

5.3.1 Awareness and attitude

Perhaps not surprisingly, the term 'clumsy' was the only term defined by all respondents. However, many expressed the view that it was no longer acceptable as a formal label for children with motor difficulties. As Miyahara and Register (2000), found in a similar survey in New Zealand, most criticisms were directed at the vagueness of the term and its derogatory connotations.

In spite of a spate of texts written specially for teachers (e.g., Portwood, 1996; 1999; Ripley et al., 1997), plus a series of campaigns by the Dyspraxia Foundation, teachers in the present sample seemed far less knowledgeable about the term dyspraxia than their medical counterparts. However, the knowledge base of any profession is likely to be a reflection of their training and attitudes to a problem. In the past, teachers were rather poorly informed about the links between movement difficulties and failure to make progress in school, and did not see children with movement difficulties as their responsibility. Although perceptions have changed over the last decade, many educationalists continue to worry that lack of coordination is a "medical" condition, which requires medical intervention.

A startling aspect of the results was the prevalence of confusion amongst teachers between 'dyspraxia' and 'dyslexia'. Some believed dyspraxia to be *"another form of dyslexia"* or "*a dyslexia of gross motor function*". In many cases, however, all that one could gather was that the respondent simply viewed dyslexia and dyspraxia as sharing membership of a fuzzy, but detrimental, set of educational difficulties.

In spite of its world-wide adoption by the research community (Fox & Polatajko, 1994; Barnett et al., 1998) only a few professionals, all therapists, were aware of the origins of the term 'DCD' and none referred to the WHO alternative, 'Specific Developmental Disorder of Motor Function'.

5.3.2 Similarities and differences in the way professionals view the terms

Awareness of a term is one thing and knowledge of its meaning quite another. Like Missiuna and Polatajko (1995), this study demonstrates that there continues to be variation in the degree of knowledge and understanding of labels. Many respondents perceived two and sometimes all three terms as inter-changeable. Such a view might have been the product of a careful appraisal of the literature. However, in most cases, the statements obtained suggested that respondents were simply guessing.

When the content of the responses was examined in detail there was agreement between professionals that all three terms were regarded as:

- Referring to movements or actions that were not smoothly performed
- Including both gross and fine motor difficulties

- As applying to children whose difficulties extended into other domains of behaviour.

There was also agreement that the term 'clumsy' referred to a milder condition than 'dyspraxia' or 'DCD'. Accordingly, 'clumsiness', unlike 'dyspraxia' or 'DCD' was never referred to as pathological. Without exception, representatives of all professions perceived these similarities. In general 'dyspraxia' and 'DCD' were seen as more severe then clumsiness, these terms were also more likely to be linked with wider problems, such as attentional problems and low self-esteem. Almost all references to cognitive difficulties were associated with the term 'dyspraxia'. In spite of these variations, however, all three terms were perceived as being linked with difficulties in school.

Differences in how the three terms were perceived could also be attributed to the individual's knowledge base. The best examples of this kind of bias were found among definitions of 'dyspraxia'. These appeared to stem from the familiarity of different professionals with the neurological or psychological literature on motor control, in which a central theme is the distinction between the planning and execution components of movement. This distinction was familiar to most therapists but was rarely mentioned by a teacher. Statements linked 'dyspraxia' to deficiencies in the planning of actions, but these varied considerably in meaning. Some used the term planning in quite a narrow sense, to refer to coordination of the components of a single purposive action. Others used it more broadly to refer to the organisation of a loosely connected series of actions.

A general problem in the study of human abnormality occurs when the purportedly neutral language in which syndromes are labelled becomes saturated with theoretical presuppositions. One example in this area, is the use of the term 'sensory-integrative dysfunction' to refer to children who have failed to acquire the motor skills expected of them. (Ayres, 1972; 1989; Fisher et al., 1991). This label enjoys currency amongst occupational therapists in the UK, some even preferring it to the terms used in this study. As a label, 'sensory-integrative dysfunction' carries with it presuppositions about the nature of the disorder and the appropriate form of assessment and remediation (Ayres, 1989). Moreover, other professionals very seldom use the term even when referring to the same children. While such differences in perspective

might be understandable within a research community, multiple terms are likely to confuse and mislead the parent or teacher unaware of the theoretical background.

Even the term 'clumsy' was not without interpretative colouration, as it often carried with it the notion of actions conducted without due care and attention. Indeed, one respondent explicitly stated that the term 'clumsy' was a label for those who *"don't concentrate on movement and actions"*. Also, the number of references to having accidents, knocking things over etc was nearly ten times higher for the term 'clumsy', than for 'dyspraxia' or 'DCD', suggesting that disability was not a causal factor in this case. This may be one reason why the word 'clumsy' in ordinary discourse is used as an epithet for a culpable action more often than a medical condition. More importantly however, statements and beliefs of this kind remind us that there may still be children in our schools today whose genuine movement difficulties are misunderstood and attributed to perversity or carelessness.

5.3.3 Implications for practice

Different professionals play very different roles in the identification of children with movement difficulties. Medical practitioners are qualified to make formal diagnoses and within allied health professionals (OT, PT, SALT) there is increasing autonomy and experienced practitioners nowadays are expected to make diagnoses within their specialist area. Teachers, however cannot make a diagnosis of dyspraxia or DCD although they may play an important role in the diagnostic process by recording their observations in the classroom. Perhaps not surprisingly, therefore, different professions emphasised different elements of the movement difficulties such children experience. For instance, most comments on handwriting difficulties were made by teachers and linked to the term 'dyspraxia'. In contrast, the majority of references to articulatory deficits and their effect on communication, although also linked to 'dyspraxia', were made by Speech and Language therapists.

As noted earlier, there remain many issues related to the question of labels and communication. However in my own experience, although professionals are becoming more aware of the problems of the child with DCD there remain issues related to identification, assessment, diagnostic labels and the implications for intervention. Without a diagnostic label children may not be able to receive intervention and often, unless a statement of Special Educational Need is given to the

child, services are unavailable. Many families trek from clinic to clinic in the hope that a diagnosis may open the door to intervention. Some children get no label and thereby lack formal identification for intervention. Other children may be given a label but it is debatable whether this influences access to and provision of appropriate services (see Chapter 6).

5.4 Conclusion

In summary, this study revealed systematic differences in how the three terms commonly used to describe children with 'specific' difficulties in the motor domain are perceived by British professionals in child health and education (Peters et al., 2001). Although it was accepted that the term 'clumsy' is out of date and unacceptable, professionals in the UK continue to use both of the alternative terms interchangeably. In the UK and in the world at large, however, there have been significant changes and DCD has gained ground. Since these data were collected the current world situation reflects that DCD is the recognised and expected term used at international conferences (DCD V Banff, 2003; DCD UK, Oxford, 2004; 2006; DCD VI Trieste, 2005; Leeds Consensus Statement, 2006) and reflected in papers and books recently published entitled 'Developmental Coordination Disorder' (Sugden & Wright, 1998; Missiuna, 2001; Cermak & Larkin, 2002; Sugden & Chambers, 2005).

A surprising finding in the present study was that references to activities of daily living were extremely infrequent. Yet, in my clinical experience, problems with dressing, undressing, eating etc are high on the list of parent's concerns. In the following chapter of this thesis a study is presented in which some of the concerns voiced by parents and carers are examined and how labels can affect service provision is examined.

Chapter 6

Study 2: Service Provision, Labels and Parent Satisfaction [2]

6.0 Introduction

Recent longitudinal studies have confirmed that children with DCD do not simply 'grow out of' their problems (Losse et al., 1991). On the contrary, without intervention the difficulties persist and are frequently accompanied by an increasing number of other problems, both at home and at school (Cantell et al., 1994; Hellgren et al., 1994; Rasmussen & Gillberg, 2000). Of particular concern to physiotherapists is a tendency for this group of children to avoid normal physical activity. This compromises fitness directly (Bouffard et al., 1996) and in turn may make these children vulnerable to future disease, including osteoporosis, cardiovascular conditions, obesity, musculo-skeletal disorders, type 2 diabetes mellitus, and mental health problems (British Heart Foundation, 2000; DOH, 2004). Although teachers share the concerns of health professionals, they are rather more concerned about things like lack of expected academic progress (often linked to poor handwriting), poor skills in PE, vulnerability to bullying and low self esteem.

Health and education provision for 'clumsy' children currently varies both within and between countries. Although recent reviews of intervention studies conclude that most approaches have a positive effect (e.g., Sugden & Chambers 1998; Pless & Carlsson, 2000: Henderson & Henderson, 2002), there is no evidence that one is more effective than the others (Ayyash & Preece, 2003). In the UK, a wide variety of therapeutic options are available through health and/or educational channels. These may be clinic, school or community-based. They may be provided as individual or group sessions, and offered on a regular basis or intermittently. For many children, however, intervention is unavailable or, at least, subject to considerable delay. One reason for this, mentioned previously, relates to labels. A label, which may be a medically accepted diagnosis or one which meets some recognised educational criteria, marks the child with a badge which often opens the door to funds that enable priority for service provision.

---

2 A version of this Chapter has been published. Peters, J. M., Henderson, S. E., & Dookun, D. (2004). Provision for children with developmental coordination disorder (DCD): Audit of the service provider. *Child: Care Health and Development, 30*(5), 463-479.

Another reason why children with DCD fail to be offered adequate intervention relates to the dearth of paediatric physical and occupational therapists. This results in high case-loads for those in the service and excessively long waiting lists. Moreover, in many localities developmental coordination disorder (DCD) is still considered to be of low priority (Dunford & Richards, 2003). In the educational system, the situation is improving but lack of expertise and uncertainty about locus of responsibility for these children continues to contribute to the uneven provision across the UK. As a result of the difficulties some parents encounter in accessing help for their children, a not insignificant proportion turn to dubious complimentary treatments, such as cranial osteopathy or unproven dietary supplementation, which usually require private funding and exert a continuous drain on parental time and finances.

As a Clinical Specialist Physiotherapist, my specific responsibility at GOSH, was to organise a service for children presenting with movement difficulties consistent with a diagnosis of Developmental Coordination Disorder (DCD). Although delivery of this service was my primary function, recent emphasis on clinical audit and evidence-based practice created an expectation that the effectiveness of the service be evaluated. The aim of the present study, therefore, was to examine parent/carer satisfaction with the service provided.

Referrals for physiotherapy assessment and intervention at GOSH come from a wide variety of sources, usually from outside the hospital but also from other departments within it. The reason for referral is the presence of movement difficulties which are thought, by the referrer, might meet the DSM-IV criteria for DCD. Sometimes it becomes apparent during the initial physiotherapy assessment that the child does not in fact meet criteria for DCD because there is a definite history of cerebral palsy, epilepsy, or other medical condition (See Chapter 2). As part of the process of deciding on eligibility for intervention, standardised tests are routinely conducted. These assessment data, tempered by practical considerations, are used to determine whether the child is offered one-to-one therapy or a home programme.

There are many ways one could evaluate the effectiveness of a programme like the one offered by GOSH. One might for example ask the children themselves, or their teachers, or seek the views of the referrer. However, at this stage, it was elected to

focus on the parents or carers of the children concerned, mainly because it was felt that they would have a view on more aspects of the service than others involved. In order to obtain as complete a picture of the families' responses to the service as possible, it was important to determine the adults' perceptions of their child's difficulties and their views of the assessment process, as well as their overall satisfaction with the intervention package. As noted in previous chapters, one of the problems often faced by parents, is a failure to obtain a clear diagnosis of their child's 'condition'. This can mean no label at all or a series of different labels, which might or might not be appropriate. This, often painful process of seeking a diagnosis, seems very likely to colour any parent/carer's view of the services they are offered. An additional aim of this study, therefore, was to build on the exploration of terms applied to these children, reported in the previous study.

In order to achieve these aims, the method of data collection chosen was postal questionnaire. This method was selected for three reasons. It provided the ability to reach the whole range of patients many of whom had travelled a long distance to attended for physiotherapy. Linked to this was low cost of data collection and processing and finally postal questionnaire ensured distance and anonymity between researcher and participants.

6.1 Method

6.1.1 Participants

The present sample was drawn from 148 consecutive referrals to an out- patient clinical specialist service at Great Ormond Street Hospital for Children (GOSH), between June 1997 and June 2000. Children were included in the study if they had attended an initial physiotherapy assessment in the out-patient department, were aged between 4 and 18 years, had no previously identified, confounding medical diagnosis affecting movement and currently attended mainstream school.

Fifty-eight children failed to meet the inclusion criteria and more details of these exclusions are given below:

- *Medical diagnosis prior to referral:* Eleven children with the following diagnoses: cerebral palsy, neurofibromatosis, metabolic condition, myopathy, postural lumbar strain, talepes, degenerative neurological condition, epilepsy, cerebral tumour, complications associated with congenital cardiac surgery.

- *Not outpatients:* two children were excluded because they were inpatients at GOSH at the time of their physiotherapy assessment. Both were undergoing investigations for psychiatric conditions thought to be, at least partially, associated with their symptoms of clumsiness.
- *Declined initial assessment offer:* Twenty-two declined to attend for various reasons (e.g., child awaiting further medical investigation, child offered assessment by local therapy service).
- *Outside age range:* Two children were outside the age range.
- *Unable to contact:* Eleven others were untraceable.
- *Pilot study:* Ten children had formed the pilot cohort.

The remaining ninety children were therefore included in the study.

6.1.2 Ethical issues

Clinical audit is part of recognised good practice and at the time when this study was carried out questionnaires for audit did not require ethical permission. The study was approved by the head of the Physiotherapy Service at GOSH to whom questionnaire responses were returned.

6.1.3 Service outline

Children referred to the present service receive a physiotherapy package comprising a comprehensive assessment, conducted by a specialist paediatric physiotherapist, a written report, hospital based one-to-one sessions and/or a home-school programme. For the majority of children assessment was completed in a single session, lasting approximately two hours.

Although the assessment battery has been expanded since this study was conducted, it has always included the administration of a standardised motor test and a series of non-standardised clinical tests and questionnaires. This test battery was the foundation upon which the assessment protocols were developed for Studies 3 and 5 reported in Chapters 7 and 9 of this thesis. Case-notes are maintained according to department policy, in line with the Chartered Society of Physiotherapy Guidelines (Chartered Society of Physiotherapy, 1995; 2000). For the present audit, data included: referral letter, an outline of the child's developmental and medical history,

scores on the Movement Assessment Battery for Children (M-ABC) (Henderson & Sugden, 1992), scores on the Developmental Test of Visual Perception (VMI) (Beery, 1989) (introduced in 1998), a sample of the child's handwriting obtained under standard conditions, a record of the clinical observations made during the assessment, copies of a sensory profile questionnaire (based on Fisher et al., 1991) which the parent/carer completed during the assessment and a copy of a physical activity and leisure interest checklist devised by the first author. For most children, IQ test results are included with the referral information.

The theoretical basis of the intervention provided can best be described as eclectic. As a paediatric physiotherapist, the researcher has obtained postgraduate formal qualifications in neurodevelopmental therapy (NDT; Bobath & Bobath, 1984) and sensory integrative therapy (SI; Ayres, 1972; 1989), both of which focus attention on the sensory-motor underpinning of movement competence. The belief that cognitive and socio-emotional factors are of equal importance reflects ideas and intervention approaches proposed by Naville (1970), Abbie (1978), Baker (1981), Stephenson et al. (1991), Henderson & Sugden (1992), Peters and Wright (1999). All of these approaches recognise that the child and family's views of the problem must be placed at the centre of intervention planning, particularly in relation to target setting. This approach also reflects the current emphasis of the Healthcare Commission formed in 2004 (previously the Commission for Healthcare Audit and Inspection CHAI) who are presently developing the Children's National Service Framework (NSF) with an expressed aim "to assess the equality of health care from the patient's perspective" (DOH, 2004).

For all families, regardless of whether they have one-to-one therapy or a home programme, the physical activity and leisure interest checklist is used to highlight the child's motivation and family 'culture'. This, in tandem, with more formal assessment data is used to link the child's interests with his or her areas of weakness. The child and family are helped to identify, with the physiotherapist, their child's strengths and specific difficulties and to work toward setting and achieving realistic goals, reviewed at follow up. Discussion of concerns raised by parents/carer/child is always encouraged. Families are urged to take exercise in order to help to promote a healthy lifestyle. Stress management through developing awareness of muscle

tension and use of breathing and relaxation is emphasised. If issues raised fall outside the province of the physiotherapist the family is directed appropriately elsewhere.

The approach taken to helping the child achieve his/her targets is also similar regardless of the mode of delivery. Ideas for achieving these targets are offered either in the form of activities to do at home, or in the form of written advice which is passed on to the class teacher, PE teacher or both, via the parent/carer. In addition, most families are provided with advice on how to help the child acquire keyboarding skills.

6.1.4 Questionnaire development

In collaboration with colleagues in the physiotherapy department, those aspects of the service relevant to audit from the parent/carer's perspective were identified. A questionnaire was then designed using methodology suggested by Oppenheim (1992). A total of 17 questions, typed onto A4 yellow paper comprised a mixture of closed, and open-ended questions (see Appendix 14). Most of the closed questions sought factual information on details about appointment waiting time, source of referral and, nested within the audit was a question aimed at gathering information about the various diagnostic labels that the child had been given. Nine common terms were included, three of which - 'clumsy', 'dyspraxia', 'developmental coordination disorder' - formed the essential core of the study reported in Chapter 5. The closed questions required forced-choice responses and were simple to quantify. For the open-ended questions, a Likert-like attitude scale was employed, in which respondents were asked to rate their satisfaction with particular aspects of the service on a five-point scale. These questions dealt with things like parents' perceptions of the home programme, its ease of implementation etc.

The 17 questions were sequenced so that the more straightforward, closed items were clustered toward the beginning of the questionnaire. Open-ended questions were placed later with the direction of scaling varied to reduce bias. Respondents were also offered opportunity to comment on their child's present progress/difficulty and leave name and personal details if they wished the Physiotherapy Service to make contact with them to discuss any concerns. The questionnaire was piloted on 10 families resulting in minor adjustments to the content.

6.1.5 Procedure

A pre-paid response envelope accompanied each questionnaire. To reduce participant bias, envelopes were addressed to the Head of Physiotherapy Department, rather than directly to the present researcher who had delivered the service to almost all of the respondents. A second mailing enclosing a further copy of the questionnaire and a covering letter was mailed to non-respondents after a delay of approximately two months.

6.1.6 Data analysis

The data were analysed using Independent-Samples T Test and Crosstabs Gamma statistic for ordinal data, SPSS Version 11 (Norussis, 2002). Wherever possible, information from respondents was substantiated by cross-checking against hospital records.

6.2 Results

6.2.1 Response rate and comparison between respondents and non-respondents

A total of 45 completed forms were returned (50%): 33 following the initial mail shot and 12 in response to a reminder. In 36 cases, the child concerned was a boy and in nine, a girl. All were attending mainstream schools.

Table 6.1 presents a comparison between responders and non-responders on relevant variables recorded in case notes. The male/female ratio was very similar, with boys outnumbering girls by 4 to 1 in both groups. The proportions of children referred via fee-paying or NHS channels was identical. There was no significant difference between the responders and non-responders on total M-ABC or VMI score. According to the case notes, a similar proportion of children had generalised motor difficulties, handwriting was of concern for more than 60% in each group and the presence of 'other difficulties' was equally common. The proportions of children attending one-to-one sessions and being offered home programmes did not differ. However non-responders were significantly ($p < .05$) older, at the time of referral, than responders and had higher verbal IQs ($p < .05$).

Table 6.1 *Comparison of Responders and Non-responders from Case Note Data*

Means, Standard Deviations or Percentages as Appropriate	Responders	Non-Responders
Gender (M/F) *n* (/%)	36/9 (80:20)	38/7 (84:16)
NHS Referral *n* (%)	20 (44)	20 (44)
Age (Months) at assessment	116 (35)	133 (42) *
Verbal IQ Score	118 (19)	129 (15) *
Total M-ABC Score	14.9 (8.8)	14.2 (9.7)
VMI Score	36.9 (26.8)	32.6 (26.6)
Provided with Home Programme *n* (%)	40 (89)	39 (86)
Attended 1:1 sessions *n* (%)	32 (71)	30 (67)

* significant at $p < .05$

M-ABC Movement Assessment Battery for Children

VMI Developmental Test of Visual Motor Integration

6.2.2 Characteristics of the 45 children whose families completed questionnaires

The children in this study were referred by hospital consultants, community paediatricians, GPs, clinical and educational psychologists, and parents. One of the first questions that respondents were asked concerned the labels which had been applied to the child over the years. Another concerned their perception of what led to the child's referral.

Diagnostic Labels. Respondents were given a list of nine terms commonly used to describe children with movement difficulty and asked to indicate any that had been used to describe their child prior to referral. Space was also provided to add any other label they had been given. Of the 45 respondents, 32 marked one or more of the nine labels. The remaining 13 wrote their responses in the open-ended box.

The results for the three labels included in the previous study were as follows: Clumsy, 4, Dyspraxia, 32 and DCD, 6 respondents. Of the remaining 'motor core' labels, 'motor learning difficulty' received the most ticks (5) with Incoordination and

Sensory Integrative dysfunction each reported as being used by 2 respondents. One person wrote 'general coordination' under 'other'. An additional 11 respondents added other labels to the list, which did not have motor symptoms as primary. These included Attention Deficit Disorder (ADD) (3); Attention Deficit Hyperactivity Disorder (ADHD) (4); Hyperactivity (1), 'dyslexia' (2), 'Tourette's Syndrome'(1), 'epilepsy'(2), 'auditory processing problem'(1), 'semantic pragmatic disorder'(1), 'developmental delay'(1), and 'OCD' (Obsessive, compulsive disorder) (1). Two respondents, whose children had/were suspected of having epilepsy did not mark any of the 'motor' boxes.

Parent/carers' perception of the presenting problem. In order to determine parent/ carers' view of why their child had been referred for a physiotherapy assessment, multiple-choice questions were employed. These included items which were viewed as primarily 'motor': 'gross motor difficulty', 'fine motor difficulty', alongside 'difficulty with handwriting' which is commonly reported to clinicians. The Questionnaire also included two items, which were concerned with associated problems that might affect motor competence (e.g., 'attention').

Although respondents were asked to answer this question by selecting "the main problem area that led to a physiotherapy referral" only 20 complied with this request. Eleven ticked two boxes, the remainder ticked three or more. In Table 6.2, therefore, parent/carers' responses irrespective of how many boxes they ticked are presented. The table shows that 22 parents/carers (51%) ticked only the "motor" boxes and 17 ticked motor plus other areas. Four failed to tick any of the "motor" categories.

Table 6.2 *'Motor' and 'Non-Motor' Problems Reported by Respondents*

Problem Domain(s)	Respondents n (%)	Mean M-ABC Score
'Motor' only	22 (51)	14.3
'Motor' plus 'Attention' and/or 'Organisation'	17 (40)	16.5
'Attention' and/or 'Organisation' only	4 (9)	9.1
Total	43[a] (100)	

[a] 2 forms incomplete

M-ABC. Movement Assessment Battery for Children (normal range = <10)

Using the children's case notes as a source, an attempt was made to corroborate the parent/carer's perception of the child's difficulties. Of the 22 children whose

parent/carer had focussed exclusively on their child's motor difficulty, 17 had M-ABC scores below the 15th percentile. In the remaining five cases, two had low VMI scores and handwriting was considered to be of major concern for all. IQ data were available for 16 of these 22 children 11 of whom had a discrepancy between verbal and performance scores (V-P discrepancy) of $\geq$ 20 points.

For the 17 children whose parent/carers were concerned about both motor and non-motor aspects of their problem, 12 scored below the 15th percentile on the M-ABC five of whom also had very slow handwriting and three also failed the VMI. Of the five children who passed both the M-ABC and VMI three were over 14 years (at the ceiling of the present M-ABC test), one scored 9.5 on the M-ABC just within the norm and one child age six years was struggling with handwriting. V-P discrepancy data was available for ten of these 17 children, seven of whom had a discrepancy $\geq$ 20.

Among the four children whose parent/carers failed to mark any of the movement related problems as a reason for referral, the notes showed that handwriting proved to be a problem for all four. One of these children also failed the M-ABC, one child was 17 (outside the M-ABC age range) and one failed the VMI.

6.2.3 Satisfaction with the service provided

Waiting time and information provided prior to first appointment. Five questions gathered information about parent/carers views of the practical aspects of the service. Table 6.3 shows the distribution of waiting times for each family along with satisfaction ratings. The majority of children were assessed within two months of receipt of the referral and all respondents were satisfied with this - although one alluded to the difficulty attending during school time. Not surprisingly, 42% of those responders who had had to wait three months or more rated this 'unsatisfactory'.

Table 6.3 *Time to Wait for Initial Appointment and Satisfaction Rating*

Time to wait (months)	n (%)	Satisfactory n (%)	Unsatisfactory n (%)
≤ 2	24 (56)	23[a] (100) *	0 (0)
≥ 3	19 (44)	11 (58)	8 (42)
Total	43[b] (100)	34 (79)	8 (19)

* significance $p < .01$

[a] one respondent did not answer this question [b] 2 forms incomplete

Parent/carer's satisfaction with the assessment and its outcome. The Assessment battery included two published standardised tests the M-ABC and The VMI. Handwriting was evaluated while the child made a drawing, carried out the VMI and completed the timed copying task. Face value of all these measures and the breadth of the assessment was given positive approval by most of the respondents. The questionnaire asked respondents how well they felt the physiotherapist had identified the child's difficulties. The battery appeared to capture the child's difficulties and helped to clarify the diagnosis and thirty-three respondents considered that this was done 'very well', 10 'quite well' and two were 'unsure'. Of the two respondents who were unsure, one had marked 'handwriting' as the only reason for their child's referral, the other 'organisation'. Cross-checking with records confirmed handwriting as the single problem domain for the first child. At assessment, the second child was shown to have difficulty with organisational skills, not considered to require physiotherapy.

Intervention options. Forty four families were offered either 1:1 sessions at GOSH in conjunction with a home-school programme, or the latter on its own. One family declined intervention of any kind opting for alternative therapy elsewhere. Thirty-one respondents attended 1:1 therapy sessions. There were various reasons why thirteen families were not offered this option (e.g., lived too far from the hospital, were unable to pay fees, found good local services). Data on the frequency of one-to-one intervention sessions attended provided by the respondents tallied well with case note data (Spearman's rho Correlation coefficient .89).

Satisfaction with One-to-one intervention. Table 6.4 shows the number of 1-1 sessions attended by the 31 families involved, along with their opinion of its effectiveness. Twenty-five respondents considered that these sessions had improved

their child's condition, six felt that there was no change. There was a significant correlation between perceived effect of physiotherapy and the number of sessions attended (Gamma = .57 p < .01).

Table 6.4 *The Relationship between Number of 1:1 Intervention Sessions Attended and their Perceived Effect Rating by Respondents*

Number sessions attended	'V. Much Improved'	'Improved'	'No change'
1	0	3	0
2-6	1	11	5
7-11	2	3	1
=/> 12	2	3	0
Total n = 31	5 (16%)	20 (65%)	6 (19%)

Gamma $p < .01$

Table 6.5 *The Relationship Between Home Programme Practice and its Perceived Effect Rating by Respondents (n%)*

Home programme Practice per week	'V. Much Improved'	'Improved'	'No change'
Never	0	3	4
Sometimes	1	5	3
1 day per week	1	3	1
2-6 days per week	3	9	1
Every Day	0	3	0
Total $n = 37$	5 (14%)	23 (62%)	9 (24%)

Gamma $p < .01$

Satisfaction with home programme. Table 6.5 shows the number of times that the child followed their home programme and their parent/carers view of its effectiveness. All but seven families followed the programme on a regular basis, albeit with varying frequency. The reasons given for not doing so included "*Hard to make X do exercises as he has year 6 SATS but we are going to try again*", "*Never, but did start exercising as recommended and joined a gym*" "*Carried out for a short time*" and "*Advice was general and local PT incorporated it into a programme*". Thirty six respondents (80% of replies) considered that the programme was clear and easy to follow, five were undecided, one was negative. Frequency of carrying out the programme correlated highly with its perceived effectiveness (Gamma $p < .01$).

6.3 Discussion

The children who arrive at GOSH for assessment are usually children whose movement difficulties are of great concern to their parents/carers. They are not children who are simply 'a bit clumsy', and their teachers are likely to have viewed them as outside the normal range for their age. In a few cases, the movement difficulties these children experience will not be rated as significant on a standardised test such as the M-ABC, but will nevertheless have a substantial impact on their everyday life. Sometimes, this is because the problem is rather specific and is not actually measurable on a test like the M-ABC. The most common example of this is the child with very poor handwriting and/or very poor organisational skills. However, there are other children who do have relatively minor movement difficulties, which have a major effect because they are linked with other difficulties or because the child is simply in the wrong environment (e.g., a highly academic school which expects rapid acquisition of handwriting proficiency).

From a series of 148 consecutive referrals to the specialist physiotherapy out-patient service at GOSH, 90 children were eligible for inclusion and for precisely half of these, a parent/carer completed the questionnaires (respondents). The richness of the data provided by the highly articulate parent/carers who did participate, more than compensated for the absence of data from non-respondents. From the information gathered, the best predictor of failure to participate was age of the child at referral, combined with the time lag between referral and the request to participate. The children of non-respondents tended to be older and to have been referred a longer time ago. In these cases, failure to respond seems likely to be a consequence of the family growing out of touch with the hospital. With the child in secondary school, they had probably become more concerned with aspects of the child's difficulties other than those provoking the original referral. In any case, the fact that the children of non-respondents did not differ from the children of respondents on either of the standardised tests of perceptuo-motor competence or on mode of therapy received, provides assurance that the respondents did not constitute a biased sample.

6.3.1 The labels

In the case of the movement-oriented labels, the data reported by parents not only confirmed the lack of consistency reported in the previous study but also mirrored the state of awareness, knowledge and opinions expressed by health and education

professionals. For example, the derogatory connotations associated with the label 'clumsy' was put very strongly by one parent who commented: *"this should be banned – being referred to as a 'typical clumsy child' did untold damage"*. The frequency with which parents in this study had encountered the label 'dyspraxia' probably stemmed from two sources. At the time of this study there was a strongly established local use of the term 'dyspraxia'. Also, many of the parents in the study were aware of the material supplied by the parent support group in the UK, the Dyspraxia Foundation. Since these data were collected DCD has become the preferred diagnostic label at GOSH. This means, of course, that any new data collected might be rather different. The fact that many respondents ticked a mixture of lay labels, such as clumsy and incoordinate, as well as DCD and dyspraxia simply confirms once again what professionals noted in the previous study.

In addition to the more motor focussed labels, some respondents noted that their child had been diagnosed as having dyslexia, attention deficit disorder (ADD or ADHD), Tourette's Syndrome and Obsessive Compulsive Disorder. As will be obvious from the discussion in Chapters 2 and 3, this did not mean that the children did not experience significant motor difficulty. Instead, their motor problems were either considered as an essential part of dyslexia or Tourette's or that the child suffered from two co-occurring syndromes. Since this study was conducted in a physiotherapy department, the assessment battery employed, correctly focussed on the motor core of the children's problem. However, if there are differences between children who come with different diagnostic labels, which have practical implications for the type of intervention, this needs to be investigated. To address this kind of question, a different approach is necessary.

6.3.2 Diagnostic issues

Both ICD-10 and DSM-IV state that a diagnosis of DCD is not appropriate if the child has a known physical/neurological disorder, such as cerebral palsy or muscular dystrophy that might be responsible for their incoordination. However, neither manual specifies a procedure for determining whether this is so. Most of the children in the present study arrived at GOSH for assessment via a long and tortuous route, attracting a range of medical opinions and diagnostic labels (see Chapter 8 case studies for examples). In contrast, many children find their way to intervention for their movement difficulties through 'educational channels' e.g., by (direct) referral to

an OT or PT from an Educational Psychologist. Such a child will probably never see a consultant paediatrician. This has implications for the application of the four criteria set out in DSM.

The contrast between the educational and medical paths to intervention echoes the distinction between DCD/Dyspraxia interpreted as a discrete syndrome in its own right, and clumsiness of movement as a symptom of another specific condition which was discussed in Chapter 2 of the thesis. The dichotomy also raises the practical question of how the principal exclusion criterion for DCD (absence of established neurological disorder responsible for the child's difficulties Criterion C) can be satisfied without involving already over-burdened paediatricians in screening all who travel the educational pathway. Although the paediatric input may not be essential for most practical purposes, the importance of differential diagnosis of rarer conditions such as cerebral tumour remain at the forefront for a few cases and does also require to be satisfied in research studies.

Apropos Criterion C, some of the children in the study reported in the present chapter had been seen previously in rheumatology or orthopaedic clinics for lax ligaments, tibial torsion, hip anteversion or unusual gait, without any specific condition being identified. Others had been investigated in muscle clinics, without clear signs of muscular pathology being found. Yet others had been medically followed-up because of prematurity and, perhaps, the suspicion of mild cerebral palsy. The diagnostic pathways followed by some of these children and the implications for the application of Criterion C are discussed further in Chapter 8.

Several children in this study were referred by psychologists, whose primary focus was on behavioural features such as hyperactivity, 'difficult' behaviour or poor social interaction. Indeed, two children whom we excluded from this study were referred from an inpatient psychiatric unit in GOSH. These children were candidates for a diagnosis of Obsessive-Compulsive Disorder (OCD), Asperger's Syndrome or Tourette's Syndrome. It transpires that many psychiatric conditions are co-morbid with DCD. Motor incoordination is also a frequently reported feature of Asperger's Syndrome (Ghaziuddin et al., 1994; Green et al., 2002), Tourette's Syndrome (Serrien et al., 2002); ADHD (Harvey & Reid, 2003) and other childhood conditions. The fact that 'co-morbidity' is the rule rather than being the exception to the rule

leads us back to the question of inhomogeneities in the mass of DCD cases (Kaplan et al., 1998; Gillberg, 2003). We can enquire whether the incoordination that often accompanies Asperger's syndrome differs from that found where incoordination is the primary or even the only symptom present (Green et al., 2002). See also Chapters 3 and 8 for further discussion.

Relevant to Criterion D, it is noteworthy that all 45 children in this study attended mainstream schools at the time of their referral. IQ scores were not available for all, but verbal IQ ranged from 91 to147, with 16 children being in the high or superior range. In all but three of the latter cases, a problem with handwriting in school was specifically mentioned by the respondent or by the child himself. Often such children read well and contribute much in discussion, but turning this into written language defeats them. Especially for those attending schools with a highly academic bias, a failure to convince the staff that the child had a specific and genuine problem requiring and susceptible to remediation, can result in substantial underachievement and disillusionment. This can, in turn, create other problems. It is profoundly unsatisfactory that such children cannot access services in many regions because their problems are not viewed as sufficiently serious.

6.3.3 Satisfaction with the service provided

One respondent's comment reflects the findings of the previous study reported in Chapter 5 in relation to lack of understanding: "*At present the service offered in the UK varies from very good ... to very poor with in my experience, some therapists having very little understanding of the condition or the impact it can have on the individual child*". Overall, the 45 parents/carers rated the physiotherapy package at GOSH very highly and had few complaints. "*We have been very lucky to be able to come to GOSH. After the very patchy and non-specific therapy offered locally it has been a lifeline in some ways*". Interestingly, the least helpful aspects of the package seemed to be the 'information and leaflets' provided. Perhaps the proliferation of information on the internet means that one of the roles of experts should be to guide parents towards recommended web-sites.

As noted above, children finding their way to GOSH are a rather complex group, sometimes difficult to diagnose for one reason or another. However, a recurring comment from parent/carer suggests that complexity may not be the only reason for

children finding their way to a Specialist Centre. The problem of waiting lists and the lack of NHS therapy provision may be just as important. "*We have waited 4 years with no prospect of an appointment*"; "*... my child is not a priority case for NHS treatment. Therefore it was important for us to find out what we can do for ourselves at home which is what was achieved*". (Note that the average waiting time for an initial physiotherapy assessment at GOSH for children in this study was < 2 months). Sometimes the remarks referred to enabling a path of care: "*We were desperate for help. GP not particularly helpful re referrals to professionals*". "*Patchy and non-specific therapy locally. The hardest part is often finding where to go for help, advice and information*". "*Education (in my experience) have overall very little understanding of the condition and in fairness to them have neither the time or resources to find out. Difficulty arises with children who fall into the borderline category – who do not qualify for special needs and hence are very much overlooked until "problems" occur*". The latter comment again reflects the continued problems of children with movement difficulties who fall in the greyer areas of medical diagnosis.

The assessment process and opportunity for clients to talk. In general, the parents/carers in this study reported a very high level of satisfaction with the extent to which the assessment identified the child's problems. In particular, respondents found the process of observing the assessment helpful and identified the opportunity to talk about the child's difficulties and their implications as the most helpful aspect of all (even in cases where follow-up by GOSH staff was minimal). One parent wrote " *we appreciated assessment from an overall view point not just his motor control. It was reassuring to talk about child's (and adult's) problem*".

The importance of observing the assessment and opportunity to talk to the therapist partly influenced the decision to allow parents/carers to be present for the assessment that is reported in Chapter 9 of this thesis. The assessment battery employed is under constant review and any new test that might be useful is carefully considered. At present, a mix of standardised and non -standardised tests are used to explore developmental history, current movement competence, problems at home and at school and a child's interests in physical and leisure activity.

The M-ABC and the VMI are two of the most commonly used tests in UK, for the evaluation of perceptual-motor function, especially in children with putative DCD. These formed the core of the assessment used here and were retained in Studies 3 and 5. In the present sample, 29 children 'failed' on either or both of these tests, suggesting that each has a useful role to play in the diagnostic process. However, the fact that 12 children passed both is also worthy of comment. As noted above, difficulty with handwriting was highlighted as a major concern for a large proportion of children in the sample, with speed of production being an additional issue for the older ones. The absence of a reliable and valid measure of legibility and speed of handwriting is a considerable problem for all professionals. In Study 5 a protocol for obtaining handwriting samples was designed and additional items were included to extend the information obtained on each child's movement competence.

One-to-one therapy and/or a home programme. There is no magical cure for DCD/Dyspraxia and no evidence to suggest that any one approach to intervention for children with DCD is better than another. Indeed, Sugden and Chambers (1998) suggest that the way intervention is delivered may well be just as important as the content. In the present study, the mode of delivery was largely determined by practical factors. Some children were able to take up the offer of one-to-one sessions plus a home programme. For others, this was not possible since the family lived far from the hospital, both parents worked etc., and the latter alone had to suffice. Since this was not a research study in which the options were carefully controlled, a comparison between families having both types of intervention and those with only one would have been difficult to interpret. Consequently, each component was treated separately. Respondents were asked to judge whether or not they thought their child had improved as a result of (a) the one-to-one intervention ,and (b) the home programme. In the first instance, the number of sessions attended correlated highly with the 'improvement' rating. In the second, a strong relationship emerged between the number of times the children practised the programme at home and its perceived effectiveness. In both cases, more than 75% of parents perceived some positive change in the child. Among the nine 'home programme only' families who reported 'no change', four had never followed the programme and three had done it 'sometimes'. For those who failed to find the one-to-one intervention effective, the pattern was less clear. In a research study it would be important to compare carefully the characteristics of those children (and their families) who did and did not respond

to the intervention programmes. There is need for randomised control trials and indeed a Cochrane Review of OT and PT for children with DCD is presently in progress (Lipson et al., 2003).

What was it about these programmes that led to their being perceived as effective? Although respondents were not asked to comment in detail, there were key elements of the programme(s) which were identical, and upon which respondents often commented. For example, the way goals were set for each child was similar in both cases; care was taken to ensure that the skills to be acquired were popular with the child and were consistent with the lifestyle of child, family and school. Whereas help with achieving these goals could be more direct in the one-to-one sessions, similar strategies were suggested to parents when this was not possible. In both cases, too, parents and children's understanding of DCD and how it affects the whole family was addressed freely with advice on how to accommodate to the problems being highly individualised. For instance, for some children, improvement in handwriting is feasible. For others, an alternative mode of communication is the only option. Such decisions are not simply dependent on the child's age and perceptuo-motor abilities, they also depend on the ethos in the school, whether parents can buy (obtain) a computer, availability of keyboarding teachers/packages, and many other factors.

The opportunity for regular (guided) practice of movement activities is something which most professionals view as an ingredient for success. To implement this principle is not always easy. Any child who has been exposed to constant failure is reluctant to keep trying. However, this is an area where experienced professionals can help parents to get the task and context right for success. Here, schools too have a role to play. As one parent suggested "*I feel that many of the exercises (in the programme) are ones that used to be playground games. I think greater awareness in education of this might encourage schools to try to introduce games back into the playground*". There is another reason why this particular group of children must be encouraged to take exercise. Recent research studies, government directives and the media all highlight the problems resulting from lack of exercise, sedentary life styles and obesity in children generally (British Heart Foundation, 2000; DOH, 2004). Children with DCD may be particularly vulnerable to these effects and end up in physiotherapy departments later in life for other complaints. Physical activity has

been shown to reduce morbidity and is now included in international health promotion guidelines (Cavill et al., 2001; Kavey et al., 2003; DOH, 2004).

There is one last question worth considering. Rather than enquire whether the children who come via the mountainous medical route to intervention are drawn from a truly different population from those who find their way via the educational route, one can ask whether the manner of arrival has implications for the design of an appropriate intervention. Likewise, one may enquire whether the intervention appropriate to the incoordination in, for example ADHD, is the same as that which is effective in 'pure' cases of DCD.

6.4 Conclusion

This study has shown that many children with DCD continue to find it difficult to obtain help for their problems (Peters et al., 2004). In some parts of the country, a clear pathway through referral, assessment and intervention has been established, but far more often parents continue to be faced with misunderstanding and delay. Moreover, there is continued uncertainty about labels in the diagnostic process and ultimately, understanding of whether DCD exists as a separate entity or is simply a symptom of numerous other childhood conditions. Some specific examples of the delays and pitfalls for both service providers and purchasers, along the route to diagnosis and intervention are presented through twelfe real life case studies in the Chapter 8.

Chapter 7

Study 3:The Difficulty of Diagnosing and Describing DCD: A Retrospective Study

7.0 Introduction

On a regular basis, children are referred to GOSH because someone is concerned about their inability to acquire the movement skills required of them. Some come from clinics within the hospital such as neurology, rheumatology or psychiatry. Others come from allied health professionals (AHPs) within and outwith GOSH and yet others are direct self-referrals. In some instances, extensive clinical notes are provided from the referring source. At other times, the physiotherapist may be the first port of call and there has been little investigation beforehand. By the end of the assessments undertaken in GOSH, however, what does emerge is a very clear picture of each child's movement difficulties and their possible causes. Although there are disadvantages to retrospective studies, the uniqueness of this database offered the potential to address some very specific questions about DCD and related disorders. Since the questions to be addressed fell into two distinct categories, one dependent on the other, analysis of the data took place in two stages. In the first stage, the questions addressed focussed on the operationalisation and application of DSM-IV criteria in relation to DCD. In the second, the objective was to re-examine the question of subtypes within a group of children who met strict criteria for DCD.

As noted earlier, neither the APA nor WHO actually specify the procedures to be used to operationalise their proposed criteria. Consequently, for every diagnostic category in the book, adult or developmental, there is a substantial literature on how this should be achieved with maximum efficiency, reliability and validity, with the area of DCD being no exception. Over the years, the department of physiotherapy in GOSH has been gradually developing an assessment battery designed to evaluate children with possible DCD in line with DSM-IV criteria. This includes two standardised, norm referenced tests- the M-ABC (Henderson & Sugden, 1992) and the Developmental Test of Visual Perception (VMI) (Beery, 1989). These two tests complement each other in that one covers a broad spectrum of motor skills/ competencies while the other has a narrower focus, providing information on hand-eye coordination only. These two tests were used in the application of Criterion A in

this study. In order to ensure that as few children as possible were missed, a lenient criterion (< 15 percentile) was adopted of including any child who failed either of these two tests. In addition to the M-ABC and VMI, a handwriting test was completed by each child. Handwriting is constantly raised as an area of great difficulty for very many of these children (74% in the study by Miller et al., 2001). At present there is no one standardised tool used internationally, and published UK norms are not available covering the age range of all the children in this study. However, handwriting speed can be recorded accurately and gives a simple, objective measure of proficiency (Barnett & Henderson, 2005).

With regard to Criterion B: the impact of the child's motor difficulty on daily life was in the first instance obtained from the referral communication. The children referred for assessment were deemed by the referrer, to have some sort of movement difficulty, which was of sufficient concern to have led to a request for specialist movement assessment. This in itself might be seen as a test of Criterion B. However, documented parent/carer and school reports, combined with talking with the child and parents during the face-to-face assessment, provided further opportunity to gain insight into the perceived degree to which day to day function was compromised. These data were especially important in relation to children who failed to meet Criterion A for 'casedness'.

Great Ormond Street Hospital (GOSH) is a world renowned Paediatric Hospital linked with the Institute of Child Health, part of London University. As such there is great expertise on the doorstep and this facilitated the application of Criterion C. The sources of information used in this study, included the referral statement along with data from formal examination(s) by the present author and other medical specialists.

The majority of the children in the study had been tested on the Wechsler Intelligence Scale for Children (WISC; Wechsler, 1992), or an equivalent, and these data were included in the case notes. Since there were no children in the study who had low IQs, examination of different interpretations of Criterion D was not possible.

In stage two of this study, the focus turns to the question of subtypes within the syndrome DCD. Intuitively, many clinicians believe that such children do not fall into one group but instead belong to a number of clearly definable sub-types.

Although a number of research studies have addressed the question of subtypes empirically, no strong evidence for even one clear subtype exists (see Chapter 2). However, there are many problems with these studies not the least of which is the failure to apply agreed diagnostic criteria for DCD. In this study, another attempt will be made to look for subtypes within a strictly defined DCD group. To achieve this objective, the technique employed in previous studies known as cluster analysis, will be used. Additionally, the question of whether the addition of those children who had a well-documented medical condition alters the profiles of performance obtained is examined.

In summary, the aims of this study were:
To evaluate one way of operationalising DSM-IV criteria, by describing the characteristics of children who do and do not meet Criteria A and C in particular.
To employ the technique of cluster analysis to determine whether children who meet criteria for DCD fall into meaningful subtypes.
To determine whether (a) the addition of children with known medical conditions to the cluster analysis alters the clusters of children which emerge, and (b) To determine whether the addition of the Pass group of children to the cluster analysis affects the stability of the clusters, which emerge. Indirectly, this process should act as a means of validating the cluster solution obtained.

7.1.Method

7.1.1Participants

The total cohort for this study comprised 157 children referred to the outpatient physiotherapy department at Great Ormond Street Childrens' Hospital (GOSH), between June 1997 and August 2001[3]. The reason for referral in each case was doctor's, parent's and/or teacher's concern regarding some aspect of the children's everyday movement function.

In order to obtain a suitable sample for the study, the following exclusion criteria were applied:
Failure to attend for formal assessment ($n = 30$).

[3] More than half of the children in this study also participated in Study 2. Data from the earlier study was used in this one, conducted two years later.

Age below six years because handwriting cannot be assessed properly at this age ($n = 8$).
Age above 13 years; outside MABC range ($n = 21$).
Incomplete records for M-ABC, VMI and/or handwriting ($n = 18$).

This left a total of 80 children, aged between 6 and 13 years with complete data sets ready for inclusion in the study. The local Research and Development Ethics Committee granted ethical approval for the study.

7.1.2 Data available

Since this was a retrospective study all data were extracted from the children's records. In addition to the initial referral letter, these included a 3-4 page typed physiotherapy report containing the following information: results and interpretation of all tests detailed below; clinical examination of muscle tone, movement patterns etc; parent-completed Developmental Questionnaire (details of birth/ developmental history including information on the results of vision and hearing tests and school progress).

Movement Assessment Battery for Children (M-ABC). The child completed all items, for his or her appropriate age band, according to the protocol in the manual (Henderson & Sugden, 1992). The test is comprised of eight items in total divided into three sections: manual dexterity (three items), ball skills (two items) and balance (three items). Total Motor Impairment Score range is from 0-40 where 0 = no impairment and 40 maximum. A cut off point at 10 represents the 15th percentile and scores > or = 10 are indicative of a possible or probable motor problem in children aged > or = 6 years.

Developmental Test of Visual Motor Integration (VMI)

The child completed the 27 drawing plates of the VMI following the protocol in the manual (Beery, 1989). The score range is from 0-27 where a higher score indicates better performance. Raw scores are converted to a standard score with a mean of 100 and standard deviation of 15.

Handwriting speed (based on a copying task)

The child was asked to copy 'The quick brown fox jumps over the lazy dog' as many times as possible in 2 minutes. The sentence was typed in 12- point font at the top of an A4 sheet of good quality white typing paper in portrait orientation. Note was made of the child's hand preference. The number of letters per minute was calculated following the guidelines of Wallen et al. (1996) who used a copying task similar to that used in the present assessment. Based on the normative data presented in Wallen's manual the 15th percentile points were estimated for children aged eight to 13 years and extrapolated for children aged six and seven years. These data were then used to identify children deemed to have below average handwriting speed ($\leq$ 15th percentile) or average handwriting speed (> 15th percentile).

7.1.3 Data extraction and synthesis

(i) *Stage one.* Scores on the M-ABC and VMI were used to define a group of children with poor motor function compared to age norms. Any child whose score fell on or below the 15th percentile on either or both of these standardised norm-referenced tests was deemed to meet Criterion A for DCD. This group of children was then sub-divided according to Criterion C. Any child diagnosed with a medical condition, with any other identified neurological pathology affecting movement, or a pervasive developmental disorder (e.g., autism) was included in a 'Medical' sub-group. This analysis yielded three groups of children; those meeting A and C ('DCD group'); those meeting A but not C ('Medical group'); those meeting neither A nor C ('Pass group'). Children in each group were then rated on the handwriting test.

(ii) *Stage two.* The DCD group was entered into a cluster analysis to search for and identify any sub-groups. Discriminant analysis, and background information from the children's records, were used to further validate and interpret the cluster findings. Further cluster analyses were performed with the Medical group added to the DCD cohort and also with both Medical and Pass groups combined with the DCD group.

7.2 Data analysis

Data were analysed using the Statistical Package for Social Scientists (SPSS version 11.0 for Windows; Norussis, 2002). Non-parametric and parametric statistics used in this study included Chi square; Pearson correlation and independent sample T-Test

and ANOVA in addition to Cluster Analysis method detailed in the following section.

7.2.1 Cluster analysis method

Cluster analysis is an exploratory data analysis tool which has been in use since 1939 (Tryon, 1939), although recent computer statistics packages have increased its popularity. It is a tool, which simply discovers structures in data but does not explain their existence and therefore reliability and validity is partly dependent on the expertise of the researcher, along with further empirical work of a different kind. The choice of variables which are entered into the cluster analysis; the cluster method chosen; the choice and meaningful interpretation of the cluster solution will all finally depend upon the researcher. Clusters can only be based on the variables that are entered into the database therefore the selection of variables must be relevant and valid to the research question. A cluster should have internal cohesion and external isolation (i.e., the variation within a cluster should be relatively small, maximal homogeneity, and the variation between clusters relatively large/greatest separation). Data may admit more than one meaningful classification e.g., books may be sorted by subject or by author.

Many algorithms have been proposed for cluster analysis, however there is no generally accepted best method (Manly, 1994, p.132). Two rather different approaches are often used and form part of the SPSS statistical package. The first known as hierarchical (or agglomerative) methods do not pre-assume a final definitive number of clusters. All objects or participants start by being alone in a group of one. A matrix of distances between all these individuals is calculated and groups that are close are merged. A dendogram, rather like the branches of a tree is produced by a step-by-step process of amalgamation until all participants form one single cluster. Within, hierarchical methods measurement of distance or closeness is performed by a variety of ways often based on squared Euclidean distances e.g., Ward; Average Linkage.

The second approach, *K*-Means clustering, involves an iterative method of partitioning. Initially, arbitrary group centres are chosen and individuals are allocated to the nearest. Participants may join or leave a group as the analysis progresses. If an individual is closer to another group's centre point than to its present group it will be

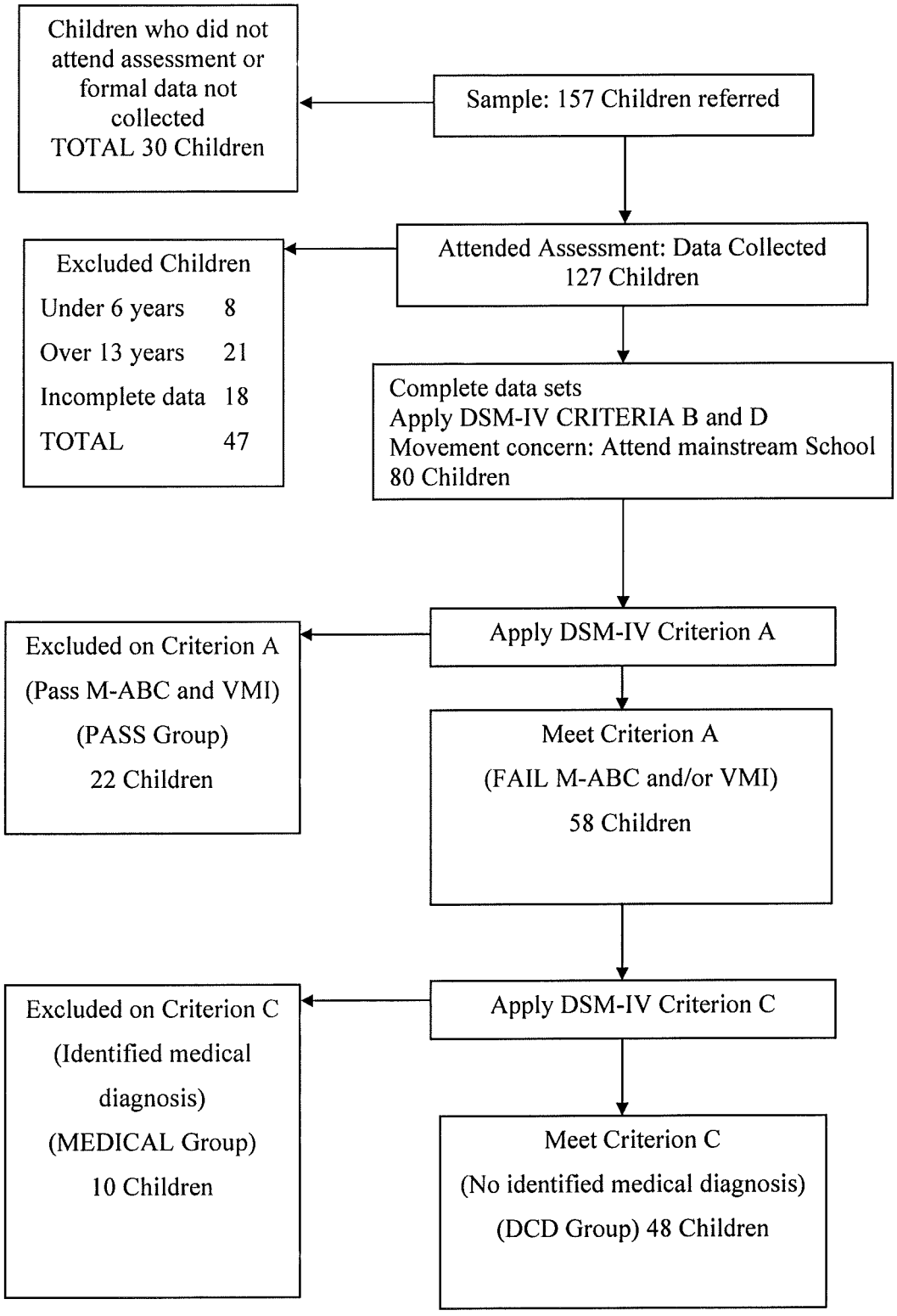
moved to the new cluster. *K*-Means clustering is particularly useful for fast analysis of very large data sets. One of the disadvantages is that the number of clusters must be specified in advance. This involves some trial and error in choosing the number of clusters. Forcing into a specified number of clusters may result in the breaking up of natural groupings or coalescence of rather dissimilar individuals.

For the present study three hierarchical cluster methods (Ward, Average Linkage and Centroid) and an iterative technique (*K*-Means) were used. These different methods were chosen to explore the data as a form of triangulation that might provide strength to the meaning of any cluster groups that appeared.

7.3 Results

Figure 7.1 summarises the procedures adopted to arrive at the three groups of interest in this study. After the preliminary inclusion/exclusion criteria had been applied, there were 80 children eligible for further consideration, of whom 63 (79%) were boys. Ages ranged from 73-158 months. (Mean = 115, *SD* = 22). All children met criterion B as previously described, and all were of normal intelligence (IQ $\geq$ 70 either as measured on the WISC and/or established from school reports).

Figure 7.1 *Flow Chart to Show the Process of Exclusion*



7.3.1 Application of Criterion A using M-ABC and VMI scores

Using the cut off points outlined (i.e., at or below the 15th percentile on the M-ABC and/or VMI), 58 of the 80 eligible children (73%) failed one or both tests. Of these 58, 23 failed both tests, 32 failed only the M-ABC, and 3 failed only the VMI. Twenty-two (28%) children passed both tests (the 'Pass group').

7.3.2 Application of Criterion C using medical records

Examination of children's records revealed that for 10 (12%) of the 80 children in the study, there was indisputable or very strong evidence to suggest that they were suffering from a known medical condition. All 10 children failed one or both of the standardised tests and thus were included in the larger sample of children meeting criterion A. Thenceforth, of the 58 children meeting Criteria A and B, the ten meeting only A, B and D are referred to as the Medical Groups and the 48 children meeting all four DSM-IV criteria are referred to as the DCD group.

The characteristics of the medical group

Details of the diagnoses of the 10 children in the Medical Group are shown in Table 7.1, along with each child's IQ, M-ABC and VMI scores. There were six boys and four girls with genetic, inherited or acquired conditions with neurological signs. None of the children in this group passed both M-ABC and VMI tests. All 10 scored below the first percentile on the M-ABC and seven (70%) also scored below the 15th percentile on the VMI. Three children scored below the 15th percentile on the handwriting copying task. Five of the six children who had a record of IQ had a V-P discrepancy > 25 points in favour of verbal score. One child had a verbal IQ of 70, at the cut off point of normal range.

Table 7.1 *Details of the Ten Children with an Identified Medical Diagnosis*

Case M/F	Diagnosis	Birth History	Total M-ABC	VMI % ile	Writing below 15th % ile	Verbal IQ	Performance IQ	IQ V-P Discrep.
71M	Phenylketenuria. Borderline average intelligence.	Premature: 32 weeks delivery.	24.5	4	Yes	70	69	1
72 F	Myopathic facies. Other family member affected. Visual impairment. Borderline average IQ.	Full term normal delivery (FTND).	32.0	4	No	Border-Line Ave.	No data	No data
73 M	Temporal lobe epilepsy	Full term Caesarean section (LSCS), failed forceps delivery.	21.5	39	Yes	120	90	30
74 M	Neurological signs: ptosis and nystagmus. Petit mal seizures. Medicated with Ritalin.	42-week delivery (cord round neck).	24.0	10	No	124	90	34
75 M	Respiratory arrest post cardiac surgery. Seizures.	Full term forceps delivery.	37.0	13	No	No data	No data	No data
76 F	Mild hemiparesis (?Cerebral Palsy).	Adopted – no birth details.	38.0	6	No	No data	No data	No data
77 F	Congenital Toxoplasmosis. Hydrocephalus + shunt. Diplegic signs (?cerebral palsy). Seizures. Partial sight.	Full term forceps delivery.	26.0	10	No	108	80	28
78 M	Non-specific dysmorphic syndrome. Pyloric stenosis. Left side neuro signs. Hearing impairment.	High forceps breech delivery.	30.0	7	No	87	60	27
79 F	Neurofibromatosis Type 1 (NF1).	FT. LSCS - failed forceps delivery.	29.0	55	No	No data	No data	No data
80 M	Benign frontal cerebral tumour	FT normal delivery	21.0	47	No	121	77	44

Normal reference ranges:
M-ABC < 10.0
VMI > 15th percentile
IQ V-P discrepancy < 15

7.3.3 Application of criterion A and C together

Although all children in the sample had been referred because of concern for their movement competence, 22 passed both the M-ABC and VMI tests. Other examinations at GOSH failed to reveal evidence of a medical condition of any kind. These children are referred to as the PASS group. Their IQ scores, scores on the M-ABC and VMI along with handwriting speed are shown in Table 7.2.

Table 7.2 *Results of the 'Pass Group' for M-ABC, VMI, Handwriting and IQ*

Case	Age in full years	M-ABC Total	VMI Percentile	Writing speed (letters per minute)	Writing below 15th percentile	Verbal IQ	Performance IQ	IQ V-P discrepancy
3	6	1.0	50	14.0	yes	91	86	5
4	7	0	55	22.0	yes	131	92	39
7	7	0	50	28.0	yes	135	85	50
10	7	5.5	37	51.5	no	123	103	20
16	7	1.5	61	5.0	yes	103	78	25
12	7	6.5	27	40.5	no	105	104	1
1	7	3.5	55	33.5	no	111	113	-2
2	8	9.0*	32	36.0	no	133	123	10
9	8	4	77	80.0	no	149	96	53
22	8	6.0	23	38.0	no	No data	No data	No data
6	8	0	95	62.0	no	153	136	17
8	9	9.5*	91	70.0	no	136	86	50
5	9	7.0	45	70.0	no	109	104	5
14	9	7.5	86	46.0	no	150	139	11
15	9	4.0	81	35.0	yes	138	97	41
11	10	9.5*	45	49.0	yes	136	90	46
13	10	7.0	73	65.0	no	121	77	44
20	10	4.0	19*	42.0	yes	120	104	16
17	12	7.0	19*	92.5	no	128	96	32
18	12	3.0	16*	114.0	no	131	84	47
19	12	8.0	50	71.5	yes	No data	No data	No data
21	12	9.0*	42	79.5	no	118	91	27

* score =/< 20th percentile

As the table shows, seven children obtained scores close to the set criterion for inclusion in the DCD group in that they scored below the 20th percentile on either the M-ABC (n = 4) or VMI (n = 3). Of the remaining 15, five children scored below the 15th percentile on the handwriting copying task. This left nine children with no movement problem at all that was detectable on the tests used in this study. Of these nine children four had V-P discrepancies in excess of 15 on the WISC. In total, 14

(70%) of the 20 children with full WISC results had a verbal performance (V-P) discrepancy of > 15 points lower on performance total score.

7.3.4 Comparison of DCD, 'Medical' and 'Pass' groups

Table 7.3 shows means and SD for the three groups on age at assessment, gender, hand preference, IQ, M-ABC, VMI, and the proportion of children who scored below the 15th percentile on the handwriting copying task. An ANOVA on age at assessment revealed no significant difference between the three groups, $F(2,77) = 2.73$, $p > 0.05$, and the proportions of boys to girls was similar in all three, with boys outnumbering girls by about three to one (Likelihood Ratio $X^2 = 2.5$, $p > 0.05$). The proportions of children who were left-handed did not vary either (Likelihood Ratio $X^2 = 1.1$, $p > 0.05$)

For IQ, the difference was significant for both VIQ and PIQ, $F(2,60) = 3.25$; $p < .05$, and $F(2,59) = 4.90$; $p < .01$ respectively. Pair-wise comparisons of the three groups then revealed that only the Medical group had significantly lower scores than the Pass group on both measures. There was no difference between the Medical and DCD groups. The three groups did not differ on verbal-performance discrepancy, $F < 1.0$.

Since the selection procedure described above had divided the groups on the basis of either their M-ABC or VMI score, an overall analysis on these variables was included to confirm group allocation. First the assumption of equal variance was examined using Levene's homogeneity of variance test. As this was significant for some of the variables (total M-ABC; Manual Dexterity and Ball skills) but not for others (VMI or Static/dynamic Balance) both parametric (ANOVA) and non-parametric (Kruscal Wallis) statistics were used to examine group differences. Both types of analysis showed significant differences between the groups on VMI, M-ABC Total Score and M-ABC Sub-section scores (min $p < .001$). Post hoc analyses using both Scheffé (parametric) and Mann-Witney U (non-parametric) were also in agreement. Comparison between DCD and Medical Groups revealed that the Medical Group had significantly poorer total M-ABC scores, ($p < .01$) significantly poorer manual dexterity (Scheffé: $p < .001$; Mann-Witney U: $p < .01$) and static/dynamic balance (Scheffé: $p < .001$; Mann-Witney U: $p < .001$). Ball skills and VMI were not significantly different.

Not surprisingly, there were significant differences between the DCD and Pass Group. This was true for all sub-sections of the M-ABC ($p < .01$ in all instances) and for the VMI (Scheffé: $p < .01$) Similarly, comparison of the Medical Group and the Pass Group showed that the Medical Group were significantly poorer on all M-ABC scores and the VMI ($p < .01$ in all cases).

Table 7.3 *Data for DCD Group, Medical Group and Pass Groups*

Variable	Pass n= 22 (Mean SD or %)	DCD n = 48 (Mean SD or %)	Medical n = 10 (Mean SD or %)	Stats. (p) Anova[b] and/or X^{2c}
Age at M-ABC/VMI Test[b]	112.7 (23.2)	112.4 (20)	129.6 (24.8)	.07
Gender Male (%)	17 (77)	40 (83)	6 (60)	.49
Preferred hand Right (%)	19 (86)	39 (81)	7 (70)	.30
Verbal IQ[ab]	126 (16.6)	120.0 (17.9)	105 (21.9)	.04*
Performance IQ[ab]	99.2 (17.4)	90.2 (15)	77.7 (11.8)	.01**
Total M-ABC[bc]	5.1 (3.2)	17.9 (6.9)	28.3 (6.0)	.00**
(1) M-ABC Manual[bc]	1.7 (1.6)	7.7 (3.8)	11.5 (2.9)	.00**
(2) M-ABC Ball skills[bc]	1.1 (1.8)	4.1 (3.3)	5.9 (2.9)	.00**
(3) M-ABC Static/dyn[bc]	2.3 (2.4)	6.0 (3.4)	11.3 (2.6)	.00**
VMI[b]	51.3 (24.2)	29.7 (23.8)	19.5 (19.5)	.00**
Handwriting speed <15 percentile (%)	8 (36)	14 (29)	3 (30)	.63

[a]IQ data available for 74% of total participants. [b]Anova [c]X^2

% for Handwriting speed, gender and preferred hand

7.3.5 Birth history

Details of the birth histories of the three groups of children are shown in Table 7.4. Over half of the DCD and Pass group were delivered by full term normal vertex delivery with no problems recorded. In contrast, only 2 of the 10 children in the Medical group were free from problems. Linked with this finding was the fact that

70% of the children in the Medical group had birth histories which suggested some type of compromise (e.g., prematurity, foetal distress or term forceps extraction). Similar problems were evident in 40% of the Pass group and 27% of the DCD group ($p < .05$).

Table 7.4 *Comparison between DCD, Medical and Pass Groups (Birth History)*

Birth History	DCD (%)	Medical (%)	Pass (%)	Statistic
Full term delivery no problems recorded	27 (56)	2 (20)	12 (55)	Chi-Square Linear-by-linear association .04
Premature ≤ 36 weeks	6 (13)	1 (10)	5 (23)	
Foetal Distress recorded ≥ 37 weeks)	5 (10)	4 (40)	3 (14)	
Forceps at term	2 (4)	2 (20)	1 (4.5)	
LSCS at term	4 (8)	0 (0)	0 (0)	
Adopted no detail	4 (8)	1 (10)	1 (4.5)	
Total	48 (100)	10 (100)	22 (100)	

7.3.6 Cluster analysis

Cluster analysis of the DCD Group

The scores of the 48 DCD Group children were prepared for cluster analysis using five variables: the three sub-scores from the M-ABC- manual dexterity, ball skills and static & dynamic balance and VMI scores, which were standardised and saved as Z-scores. Handwriting scores (letters per minute for the copying task) were standardised, and standardised residuals were calculated in order to adjust for age effects. Since the largest correlation between any pair of these variables was .53, one can be fairly confident that each represents a slightly different component/dimension of motor performance.

Table 7.5 shows that cluster analyses using the Ward, Average Linkage, Centroid and K-Means methods all produced a 4-cluster solution with between 93.8% and 100% correct membership predicted. The Ward method produced a 4-cluster solution with 100% correct classification of the original grouped cases. Discriminant analysis for a 5-cluster solution was slightly lower at 97.9% accuracy. Average Linkage gave 97.9% correct for a 5-cluster solution compared to 93.8% correct classification for a

4-cluster grouping. A further method (Centroid) produced 100% correct membership on either 4- or 8-cluster groups however there was wide spread and some of the clusters contained three or fewer children. A contrasting iterative method (K-Means, Quick Cluster) suggested four clusters. For consistency with the majority of studies reviewed in Chapter 2, the Ward method was chosen as the principal method for the present study. Comparisons using Cross-tabs (Chi Square: Eta for Nominal by Interval data) between Ward and K-Means; Ward and Average Linkage (4 or 5 group solution); Ward and Centroid (8 solution) were all significant at $p < .01$. However Ward by Centroid 4-cluster was not significant.

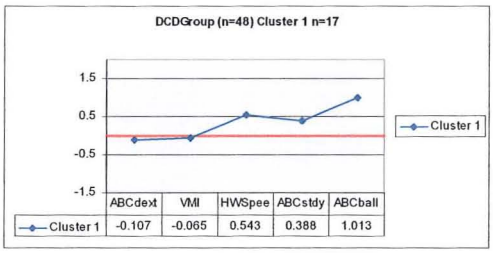
Table 7.5 *Cluster Analyses Results of DCD Group (N = 48 children)*

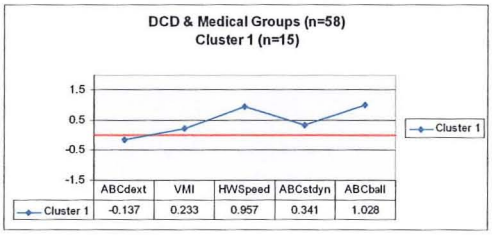
Cluster Method	Clusters (*n*)	Number of children in each Cluster Group (*n*)	Discriminant Analysis. Correct predicted group membership (%)
Ward (Agglomerative)	4 5	17, 13, 9, 9 14, 12, 9, 8, 5	100 97.9
Average Linkage (Agglomerative)	4 5	25, 17, 4, 2 17, 16, 9, 4, 2	93.8 97.9
Centroid (Agglomerative)	4 8	42, 3, 2, 1 15, 14, 9, 3, 3, 2, 1, 1	100 100
K-Means (Iterative)	4	17, 13, 10, 8,	100

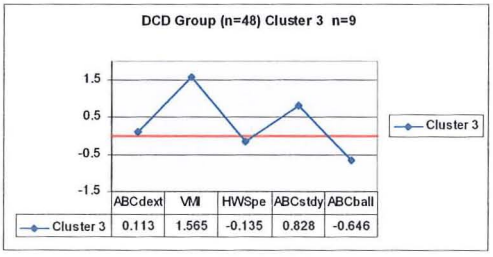
Results of the Ward 4-cluster solution gave distinct cluster groups which differed significantly on all the five variables ($p < .01$), M-ABC Manual dexterity, Ball skills, Static & dynamic balance, VMI and Handwriting standardised residuals. Three significant functions were identified and the highest correlations between each variable and any discriminant function were: Function 1. Static & dynamic balance (-.42); Function 2. Ball skills (.72) and VMI (.60); Function 3. Manual dexterity (.59) and Handwriting standardised residual (-.56).

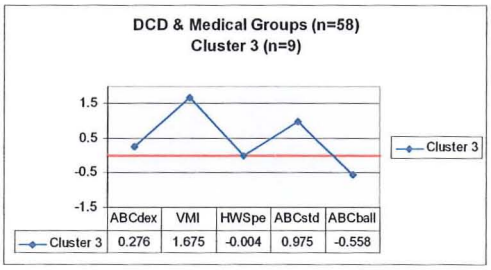
The four different cluster group profiles are shown in Figure 7.2. Note that scores for each variable are given in Z scores plotted in relation to the group ($n = 48$ or $n = 58$) mean. Details of the clusters are given in the following section.

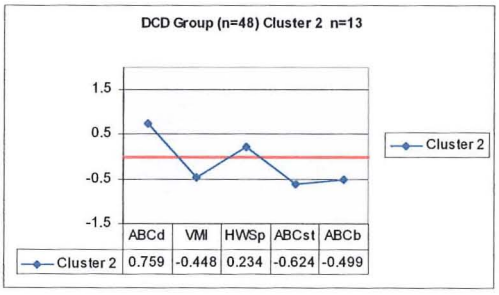
Figure 7.2 *DCD (n = 48) and DCD+ Medical Group (n = 58) Clusters*

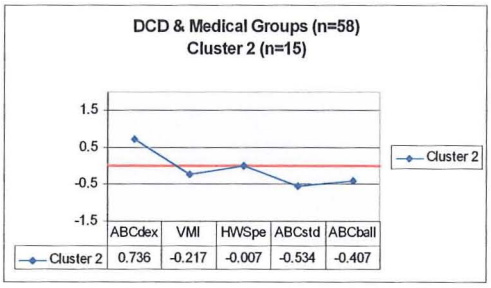


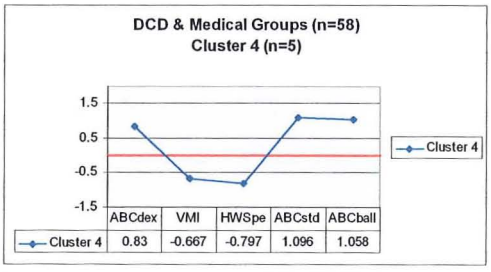


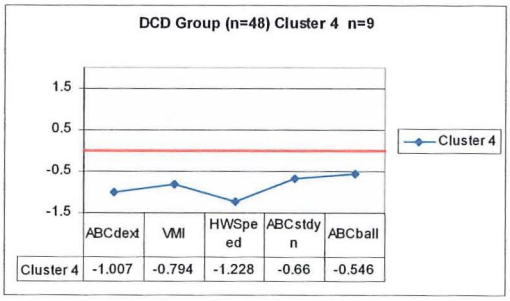


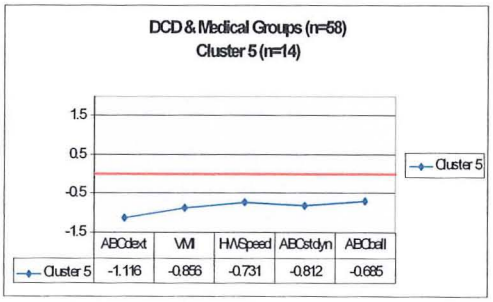












Cluster 1 the largest group (n = 17) performed comparatively above the total group mean on Ball skills (+1.0) and Handwriting speed (+0.5).

Cluster 2 (n = 13) were below the total group mean on ball skills (-0.5), balance (-0.6) and VMI (-0.5) and above the mean for dexterity (+0.8)..

Cluster 3 (n = 9) were below the total group mean on ball skills (-0.6) but above the group mean on balance (+0.8) and +1.7 SD above the mean on VMI.

Cluster 4 (n = 9) were at least .5 SD below the total group mean on all scores with manual dexterity (-1.0) and handwriting (-1.2) where they performed least well.

Cluster analysis on the combined DCD and medical groups

Results of a further cluster analysis of combined DCD and Medical groups (n = 58) produced a 5-cluster solution with 100% of the original grouped cases correctly classified. Original DCD group clusters 1 – 3 showed only small changes when re-clustered with the addition of the 10 Medical Group children. The fifth cluster group was made up of 60% of the Medical Group and seven of the DCD Cluster 4 children. A new cluster was identified with VMI and handwriting scores below the group mean in contrast to all M-ABC scores above the group mean.

Cluster 1 (n = 15) Similar to Cluster 1 DCD (Contained one Medical group child).

Cluster 2 (n = 15) Below the mean on ball skills (-0.4) and balance (-0.5) (Contained 3 medical group children).

Cluster 3 (n = 9) Remained identical to DCD Cluster 3 (Contained none of the medical group).

Cluster 4 (n = 5) Below the mean on VMI (-0.7) and Handwriting (-0.8) but above the mean on Dexterity (+0.8), Ball skills (+1.1) and Balance (+1.1) (Contained no medical children).

Cluster 5 (n = 14) The profile of this group was similar to DCD Cluster 4, all scores lay below the group mean with Manual Dexterity (-1.1) the worst area. This cluster

contained six medical children alongside seven of DCD Cluster 4 mentioned previously (the lowest performing group). One further child had moved into the present Cluster 5 from the original clustering of the DCD group.

Cluster analysis of combined DCD, Medical and Pass groups

Introduction of the Pass group to the combined DCD and Medical cohort (n = 80) again suggested a 5-cluster solution with 93.8% correct classification. Twenty-one of the Pass group (95%) clustered together in Cluster 1 (n = 28). Cluster 2 (n = 12) contained mainly DCD children with just one Medical group child and one Pass group child. Cluster 3 (n = 15) contained 14 DCD and one medical child. Cluster 4 (n = 9) remained identical to Cluster 3 previously mentioned and contained the same nine DCD children. Cluster 5 (n = 16) was made up of eight Medical group children and eight DCD children.

7.4 Discussion

This retrospective study focussed on a group of children referred to a specialist paediatric hospital because of concern for their movement and coordination. Retrospective data analysis relies on accurate records and is prone to bias in recall or selection. However, the physiotherapy department at GOSH has a strict protocol for writing and storing medical records, and it is my belief that the data employed in the study were as objective as data collected prospectively would have been. Unlike many studies in the field, background history including reports of medical examination was well documented.

A major problem to be dealt with in many retrospective studies, is the effect of attrition. Although the 80 participants in this study were selected from a total cohort of 157 children, this was not actually due to attrition but was the result of strict application of criteria designed to ensure that completed comparative data were available on all the participants. For example, some children were excluded early on because they had not undergone full assessment at GOSH, part having been done elsewhere. Others simply failed to attend the appointment offered. Some children were outside the age range for the M-ABC and it is unfortunate that the forthcoming 2nd edition of this test battery, which extends the age range above 13 years, was not available at the time of this study. The referrals were spread over a period of years and the VMI was not used in the physiotherapy department until 1998 accounting for

incomplete data sets. None of these factors seem likely to have affected the outcome of the study.

7.4.1 How satisfactory were the chosen criteria?

The first objective in this study was to examine the effectiveness of one way of operationalising the criteria set out in DSM-IV for DCD. At present, the APA offers no definite guidelines as to how to operationalise any of their proposed criteria, although research editions of the manual suggest two-standard deviation below the norm as a cut point, when a standardised test is being used. For conditions other than DCD there are sometimes better guidelines with quantitative criteria provided e.g., for ADHD a specified number of listed symptoms must be present, but for DCD debate about how best to achieve reliable classification continues (Henderson & Barnett, 1998; Geuze et al., 2001). In this study, all of the children had already been referred to physiotherapy because of concern about movement difficulty so close examination of Criterion B was not possible. The main focus, therefore, was on Criterion A and C.

Unlike many of the studies reviewed earlier, the schools attended by all of the children in this study were known and IQ was recorded for most of them. All attended mainstream schools and there was no confirmed report of learning difficulty in any child. In fact, only one child scored 70 on the WISC, right on the cut- point for moderate learning difficulties (-2SD) and 19 were actually very bright indeed (VIQ =/> 130, i.e., +2SD above the mean). The children came from reasonably similar backgrounds, without socio-economic deprivation, and a fair proportion attended highly academic schools, often in the private sector. Although I have elected not to focus attention on the application of criterion D in this thesis, and the discrepancy notion in particular, all of these children would actually be considered of average IQ or above and most would show a large discrepancy between their motor and cognitive ability, however it was measured. In many cases, the child's motor impairment appeared to be very out of line with their cognitive ability. To give a few examples, in the DCD group, there were seven children who had verbal IQs 2 SDs above the mean and M-ABC scores 1.75 SDs below the mean (i.e., below the 5th percentile). Thus the discrepancy between cognitive and motor function was approaching 4 SD. In the medical group, three children fell above 1 SD for VIQ and

had M-ABC scores on the 1st percentile (> -2SD) – a discrepancy of three SD. (See also case studies in Chapter 8).

To operationalise Criterion A, two tests, the M-ABC and VMI, were chosen to represent standardised measures and cut-off points employed in the way adopted by Jongmans (1993), i.e., the child was deemed to meet Criterion A if s/he fell below the 15th percentile point on either or both tests. From a total of 80 children, this method correctly classified 58 of the 80 participants, i.e., 73%. Geuze et al., (2001) recommended that the M-ABC 5th percentile be adopted in research studies and had this criterion been adopted in the present study, the number correctly classified would have fallen to 49 (61%), (48 with M-ABC at or below the 5th percentile and one child above the 15th percentile on the M-ABC but below the 5th percentile on the VMI). At first sight this might seem that the chosen tests were not as effective as one had hoped but there were reasons for this, which will be discussed below. As a practical point, in clinical practice, a more lenient criterion may be suitable in order to capture and identify potential movement difficulties that might have a greater impact and affect on the child in the future.

In many investigations, all of the 58 mainstream children failing the M-ABC/VMI in this study would have been classified as DCD. However, what this study reveals very clearly is that this would have been incorrect in 10, i.e., 17% cases, if one interprets Criterion C very strictly and has the facilities to do so. Put the other way round, of course, the findings indicate quite clearly that 'pure' DCD does indeed exist! A highly specialised hospital provides the facility to identify a wide range of medical conditions that in other non-clinic based studies may not come to light. In this study, an experienced physiotherapist's clinical observations were recorded in the notes and complemented the medical examinations and diagnoses provided by doctors. The screening for Criterion C therefore usually drew on two independent opinions. When non-medical e.g., education and psychology professionals try to identify children with DCD there is no way that they can address Criterion C reliably. Geuze et al. (2001) highlight the inconsistency and lack of strict adherence to DSM exclusion criteria.

As noted above, 10 children in this study, who met Criterion A would not have met criteria for DCD as all were excluded by their medical condition. Although DSM

provides some examples of what constitutes a medical condition (CP, DMD) the implication is that there should be no definite neural or muscle pathology. The medical group comprised eight children with abnormal neurological findings (two who some would have classified as having cerebral palsy), plus one child with a complex picture including symptoms of myopathy and a child with phenylketenuria, which affects early nerve cell maturation. Two children might have been viewed as displaying the typical 'clumsy child syndrome' but each had actually had subsequent investigation which revealed underlying medical diagnoses (NF1 and benign tumour). In the former case, it is unlikely that the diagnosis would be passed on to educational professionals. Although, none of the medical group children would have stood out as looking abnormal as they moved around their mainstream school, the M-ABC however showed that all 10 scored below the 1st percentile on the M-ABC and 60% scored at or below the 10th percentile on the VMI, suggesting that this group of children were at the severe end of the impairment continuum. The two children who were very clearly on the borderline between CP and DCD and might best have been described as having mild cerebral palsy (diplegia and hemiplegia) both scored very poorly on the M-ABC and VMI but surprisingly did not seem to have very slow handwriting (both scored above the 15th percentile). This may reflect the comparative non-involvement of the handwriting arm since each child had elected to write with the more functional hand. Cerebral palsy as discussed in Chapter 2 is not a clear-cut diagnosis and many believe that there are always children who fall in the fuzzy borderland between cerebral palsy and DCD. Neither of these 'medical' children presented as severe CP and each chose to see herself as 'normal' and in no way educationally or medically different and it is very unlikely that teachers would perceive them as suffering from CP.

Several children in this study 'passed' both tests. Since all of the children tested had been referred because of concern about their movement, the obvious questions that follow are: (i) did these children really have no problem?, (ii) was there something wrong with the criteria chosen, (iii) did the two tests fail to capture aspects of motor function that may lie in other domains such as cognitive planning or behaviour.

Before one can conclude that the children did not have a problem it is pertinent to examine the criteria selected and some characteristics of the tests chosen. Seven children in the 'Pass' group came close to failing Criterion A as they did in fact fall

in the grey borderline area between the 15th and 20th percentile on either or both the VMI and M-ABC. Variation of scores around the cut-points in the M-ABC manual are unfortunately not expressed in terms of a standard error of measurement and this may have influenced the grouping of a few of these children who lay near the boundary. Sliding the criteria either up, as suggested clinically, or down as recommended for research would have captured more or fewer of the 'Pass' group. Turning to the possibility that the tests chosen lack sensitivity, there are one or two reasons that might account for children with a problem not being identified. The M-ABC allows practice trials, which children may 'fail' before they subsequently pass formal trials. Dependence on practice trials to succeed may indicate difficulty in attention, planning or organising novel tasks. In my experience, there are children like this who only pass because of the practice trials, and these may be children who have more problems with everyday tasks which are novel. Another factor, which may account for the existence of the Pass group, is that the M-ABC (and VMI) was administered in a quiet, distraction-free room, one-to-one with the physiotherapist. A child may pass an assessment test under 'ideal' conditions but problems may become apparent in other settings, such as at school, when the environment around the child is usually turbulent and neither one-to-one nor distraction-free. It is essential to address the ecological validity of the assessment and consider whether the Pass group would have failed under different environmental conditions. As far as the usefulness of the VMI is concerned, which does not allow any practice, very few children who do not fail the M-ABC are picked up by this test (in this study only three). However, it may be that these few children are in fact different from the others in terms of the pattern of their performance and its underlying cause. After the present study was conducted, a new edition of the VMI (Beery, 1997) was published which offered the facility to examine separately the visual perceptual and motor components of the copying tasks, which comprise the main test. Consequently, this allows us to differentiate between the child whose copying difficulty may be caused by the fact that he/she cannot 'perceive' the characterstics of the shape to be drawn from the child whose visual perceptual abilities are intact, but who cannot turn perceptual input into accurate motor output. In Study 5, the question of whether and how the VMI should be used in the diagnostic process will be addressed further.

Many children are known to have handwriting problems and DSM-IV specifically mentions this as one manifestation of DCD in older children. Handwriting

assessment was not used to determine group membership, in the present study, as a reliable test with norms across the age range, on a par with the M-ABC and VMI, has not yet been published. However, handwriting speeds can be recorded accurately and give a simple, objective measure of proficiency (see Barnett & Henderson, 2005 for a review). Handwriting speed was standardised for age and included as one of the variables in the cluster analyses. Additionally, the children's copying speed was compared with published data (Wallen et al., 1996) with approximate 15th percentile cut-off point calculated for children aged from six to13 years to give an estimate of 'slow' handwriting. This helped to further describe the Medical, DCD and Pass groups. Of the 22 children who failed to meet Criterion A, as it had been applied, eight were below the 15th percentile on this handwriting task. Thus all but nine children (89%) referred were successfully identified using a combination of a movement test (M-ABC), a perceptual-motor test (VMI) and a graphomotor test (handwriting).

Seventy-nine percent of children in this study showed a V-P discrepancy on the WISC of 15 points or more. Interestingly, this proportion comes close to the 76% of children from the Newcastle Study detailed by Gubbay (1975, p. 87). Thirty years ago a discrepancy of this order was almost taken as a diagnostic criterion for 'DCD' and in some small pockets of the UK, this idea lives on. One of these pockets formed a referral source for the children in this study and may account for the high proportion of children with this characteristic. Although the significance and use of a V-P discrepancy continues to be an issue in the field of developmental disorders (Henderson & Barnett, 1998), this group of children probably do need further investigation, as they may be different from children whose verbal and non-verbal cognitive abilities are similar.

Examination of any of the literature which focuses on the V-P discrepancy almost invariably reveals an allegiance to the term 'dyspraxia'. All the children in the present study had initially been referred as showing either putative DCD or dyspraxia. Whereas a child would not meet criteria for DCD in the presence of a medical diagnosis no such criterion applies for the identification of dyspraxia in the UK, which is interpreted by many to mean no more nor less than difficulty planning movements (however that may be interpreted!). As this study was hospital-based the term 'dyspraxia' was possibly used, for some children in the medical group in

particular, because of behaviours which really did resemble those shown by individuals with known later onset pathology (see Chapter 1). In other studies, especially by occupational therapists and speech and language therapists, tests of gesture and sequencing are often included as indicators of dyspraxia (Hoare, 1994; Dewey & Kaplan, 1994; Hill, 1998; Macnab et al., 2001; Green et al., 2002). Objective gesture and sequencing tasks were not included in the present study as these were not part of routine physiotherapy clinical assessment at GOSH, although informal clinical observation did include imitation of postures and action sequences.

7.4.2 What does this study tell us about subtypes?

The variables included in any cluster analysis should be as different as possible, as variables that are too similar are likely to confound each other. Also, a set of variables that is too close in 'action space' is unlikely to reveal any of the dissociations that are of most interest. In this study, choice of variables was somewhat limited because the study was retrospective and the test battery was not designed with subtyping in mind. However, the three sub-sections of the M-ABC, providing measures of manual dexterity, catching/aiming, static and dynamic balance, along with the VMI and speed copying seemed to offer a reasonable breadth, as well as some opportunity for comparison with earlier studies.

As noted in Chapter 2, two of the six studies reviewed were very different from the others, Myahara's (1994) because of the sample used (Miyahara, 1994) and Wright & Sugden's (1996a) because of the type of measure employed. These two studies are not comparable to the present one and will not be considered further. The remaining four studies (Jongmans, 1993; Dewey & Kaplan, 1994; Hoare, 1994; Macnab et al., 2001), however, were comparable in that all included a measure of static balance and of manual dexterity, and the VMI was used in three of the four.

The cluster analyses undertaken on the children with 'pure' DCD in this study identified four clusters, all with significantly different profiles. In numerical terms, this outcome replicates the above-mentioned studies but is of no consequence really as the number of clusters is often 'selected' by the researcher. In the present study, an (indirect) attempt was made to validate the cluster solution by examining the stability of the DCD cluster solution obtained, when the 10 children with a medical diagnosis were added to the analysis and then the 22 'Pass' children. When the 'Medical

group' were added the four cluster solution was replaced by a five cluster solution but encouragingly the DCD clusters remained relatively stable with almost all of the medical group joining the poorest DCD group and only seven children moving group. Similar cluster stability was revealed when the 22 Pass children were introduced with 21 of these children clustering into a group, on their own.

The 48 children with DCD entered into the analyses fell into four groups, two which showed rather even or flat profiles and two which were markedly uneven. Cluster 1, the largest cluster, contained 17 children whose performance was average or above average on all tasks compared to the group mean. This finding is not unique. It would seem that any sample of children with DCD will contain children whose profile is even across **all** variables, regardless of what is being measured e.g., Jongman's Cluster 1, and Hoare's Cluster 2 also showed this pattern. Similarly, in most studies, we find a group that is much poorer on all variables (e.g., the one described as Cluster 3 by Macnab, which replicates that of Hoare, 1994). In the present study there were nine children whose difficulties were marked and this number increased when the medical group were added. Clinical notes supported the idea that many of these children had shown early signs of possible damage to the neuromotor system at or around the time of birth. Whether these characteristics such as premature birth actually preclude the diagnosis of DCD is a separate question much debated.

Two groups of children with markedly uneven profiles emerged in this study. In one group (Cluster 2), **poor balance and ball skills** contrasted with good manual dexterity, average handwriting speed and **slightly below average VMI**. This group can be compared to the second cluster described by Macnab et al. (2001), as replicating that of Hoare (1994) (except that the children's VMI scores were better in the latter studies). In the other uneven cluster emerging in this study (Cluster 3), **poor ball skills, was accompanied by good balance**, average manual dexterity and handwriting speed and **above average VMI scores.** This last group was similar to Jongman's (1993) Cluster 5 who also had poor ball skills, alongside good balance. How might we interpret the difference between our two profiles? First, it might be worth noting that if any finding is consistent across **all** cluster studies, it is the fact that some children have good balance and others have poor. Since postural control underpins all movements, large or small, the origins and implications of this variation is definitely worth pursuing. Since both of our groups of children were poor

at ball skills, we might look, for example, for differences in **how** these two groups approach the catching task. Another finding within cluster studies is that the VMI has a role to play in distinguishing one group from another. In our two groups, one was above average on the VMI and the other below. Although it is hard to see how copying a static figure relates to catching a moving object, further investigation of the differences in **how** children who pass and fail the VMI approach catching might be productive. In the end, however, it must be conceded that this kind of speculation is very post hoc. Whereas one researcher might focus on one contrast between variables that differentiate between two clusters, another might choose something quite different. Also, if other variables had been entered into the analysis, the picture might change again.

Macnab et al. (2001) provide an excellent discussion of the problems to be faced when interpreting cluster studies. These will not be repeated here. As a result of their deliberations they come to the conclusion that "cluster analysis does have a role to play in studies designed to increase our understanding of the aetiology and treatment of DCD (p. 69)". In the light of the continued problems with population definition, diagnosis, variable selection etc, however, the use of cluster analysis will not be continued in this thesis and the question of whether children with DCD show specific patterns of impairment will be addressed in a different way.

7.5 Conclusion

This study examined 80 children referred with possible DCD. A version of DSM-IV criteria were used to group the children according to whether they met criteria for the condition in question. The study showed that the tests selected to operationalise Criterion A yielded a 70% success rate - the M-ABC alone identified almost 70% of the children and combined with the VMI identification increased to 73%. The rigorous exploration of medical records to operationalise Criterion C showed that within the referred group a small number did have a definite medical condition, which carried increased vulnerability to impaired motor function. These children may not be identified unless careful medical screening has been undertaken.

A significant number of children thought to have a movement problem were not identified as having DCD. It is possible that this may reflect inappropriate referral (Dunford & Richards, 2003; Dunford et al., 2004) but it seems more likely that these

children's movement difficulties were either not picked up by the two standardised tests used, or were different in their origin. For example, a child who is genuinely 'clumsy' in every day life because of lack of attention might not fail the Movement ABC. Thus, the referral was reasonable, as was the result on the M-ABC. More than one third of this group had slow handwriting. Future studies should consider not only motor performance on tests such as the M-ABC and VMI but also reliable standardised handwriting assessment is needed. It was not possible in the present study to explore potential compromising abnormal anthropomorphic features or screen for the presence of co-occurring conditions such as behaviour problems, dyslexia, speech and language impairment or children conceivably on the autistic spectrum. If DSM-IV criteria are to be rigorously operationalised, then a much broader assessment protocol is required so that both the core motor problem and any associated/co-occurring problems can be documented.

Chapter 8

The Value of the Single Case Study: What Real Life Examples Can Add to our Knowledge of DCD

8.0 Introduction

The type of research question posed will determine the methodology chosen. In the present chapter, single case studies are used for two purposes. First they are used to bring to life some of the theoretical and practical issues raised in previous chapters. In other words the experiences of 'real' families are described in such a way that the problems still to be solved by researchers are placed in the context of everyday life. Second, it is intended that the stories of individual children will complement the more general findings reported in Studies 1 and 2.

The case studies are presented in five groups, given the broad titles:

1. DCD - does it exist as a syndrome?
2. The pros and cons of labels.
3. The application of DSM-IV Criterion A: handwriting as a special problem.
4. The application of DSM-IV Criterion C: signs and symptoms of medical conditions.
5. The application of DSM-IV Criteria: Dealing with co-occurring diagnoses.

Within each group the children described, have been selected to illustrate a different aspect of the broader issue addressed. For ease of reading, a similar format is adopted. Following a brief background statement, the child's movement difficulties are outlined with particular reference to when the difficulties were noted and by whom. In the next section, focus moves to the more formal aspects of the diagnostic process, including the type and timing of the assessments used to make decisions as well as the various professionals involved. Last, but not least, the impact of the child's difficulties on everyday life at home and at school are described.

Although not rigidly adhered to in terms of order, it should be obvious to the reader that the material within the case studies permits discussion of the application of DSM-IV criteria. Broadly speaking, the first three sections in each case study relate

to Criteria A and C, and the last section exemplifies Criterion B. In the comment, at the end of relevant case studies, therefore, pertinent points concerning the application and interpretation of these criteria are included. Since all of the children in this study are of average or above average intelligence, no further mention is made of Criterion D.

A series of 12 case studies is presented. Summary data provided in Table 8.1, show that all of the children were of school age, and boys outnumbered girls by 3:1 (9 boys and 3 girls). Although each child is different and has a different story to tell, in every case, movement difficulties in the presence of average or above average intelligence is a defining feature. All the case studies are taken from the records of real children but with names and any features that would identify them altered or omitted. Parents and children were consulted regarding the use of data and anonymity was assured.

Tables 8.1a-e Summary Data on 12 Children Described in this Chapter.

Table 8.1a *DCD - does it exist as a syndrome?*

Case name Referral age	Case Number and Title	M-ABC (Percentile) VMI (Percentile)	IQ (Standard Score) Verbal (VIQ) Performance (PIQ)	Key Points
John 6 years	1). History repeats itself- a 'classic case'.	M-ABC 6 yrs:Total: 8 M-ABC 11 yrs Total: 1 VMI 6 yrs 70 VMI non-motor 68	VIQ 128 PIQ 112	Classic 'clumsy child syndrome' Resembling reports by Walton et al. (1962) Persistent handwriting problem M-ABC score changes - impact greater in Secondary school. Many professionals involved
Frank 8 years	2). 'Pure' cases do exist	M-ABC 8 yrs Total 7 10:10 yrs 11 VMI 8 yrs 39	VIQ 117 PIQ 96	'Pure' DCD with movement problem but no perceptual, attention or behaviour difficulty
Annie 8 years Followed from age 8 to 16 +	3). Not all children with DCD are boys	M-ABC 10 Yrs Total 1 All, but manual worst VMI 10 yrs 8 VMI 13yrs 14	VIQ 111 PIQ 77	Girls do exist with marked DCD Label 'clumsy' caused distress Persistent problem beyond secondary school

Table 8.1b *The pros and cons of labels*

Case name Referral age	Case Number and Title	M-ABC (Percentile) VMI (Percentile)	IQ (Standard Score) Verbal (VIQ) Performance (PIQ)	Key Points
Kevin 11yrs	4). A label can be a great relief.	M-ABC 11 yrs. Total 1 All sections VMI 16	VIQ Above average	Diagnosis took many years – relief when label given. 'CP'/DCD fuzzy boundary? 34 week twin history
Adam 5 yrs	5). Negative aspects of labelling.	M-ABC 5 yrs Total 1 Manual dexterity and balance worst VMI 77 VMI Non-motor 95	Above average. VP discrepancy 40 points	Labels: ADHD, dyspraxia, gifted, DCD by age 5 yrs! Investigations via several specialists before diagnosis Ehlers Danlos Syndrome OT waiting list > 2 yrs.
Brian 9 yrs.	6). No label can be disastrous.	9 yrs. M-ABC & VMI not tested Gross motor only	Average IQ	Clumsiness that was serious but unrecognised. Gross motor skills poor, but fine motor ok. Teased and bullied – late diagnosis of muscular dystrophy.

Table 8.1c *The application of DSM-IV Criterion A: handwriting as a special problem*

Case name Referral age	Case Number and Title	M-ABC (Percentile) VMI (Percentile)	IQ (Standard Score) Verbal (VIQ) Performance (PIQ)	Key Points
Gerald 7yrs	7). How do we fit handwriting problem into the broader picture?	M-ABC 7 yrs Total 84 VMI 7yrs 61 VMI Non-motor 50	VIQ 103 PIQ 78	Sequencing difficulty not captured by M-ABC Visual perception and behaviour good Reading, writing, gesture poor

Table 8.1d *The application of DSM-IV Criterion C: signs and symptoms of medical conditions*

Case name Referral age	Case Number and Title	M-ABC (Percentile) VMI (Percentile)	IQ (Standard Score) Verbal (VIQ) Performance (PIQ)	Key Points
Peter 7 yrs	8) Childhood arthritis and DCD – separate the problem.	M-ABC not tested VMI not tested	Average IQ	Poor motor function associated with morning joint pain & stiffness. Fails to meet Criterion C. Referred to rheumatologist Health screen important.
Michael 11 yrs.	9). Chicken and egg. DCD or brain tumour - which came first?	M-ABC 11 yrs. Total 1 Ball skills normal VMI 47 VMI Non-motor 74	VIQ 127 PIQ 82	Fails to meet Criterion C? Child with DCD may also develop a brain tumour – cause or effect?
Mary 10 yrs	10). NF1 – an under-diagnosed condition	M-ABC Total 1 Dexterity normal balance poor. VMI 10 VMI Non-motor 88	Average IQ	Late diagnosis: labelled delayed motor development, dyslexia, clumsy, dyspraxia. NF1 underdiagnosed, health screen important.

Table 8.1e *The application of DSM-IV Criteria: Dealing with Co-occurring diagnoses*

Case name Referral age	Case Number and Title	M-ABC (Percentile) VMI (Percentile)	IQ (Standard Score) Verbal (VIQ) Performance (PIQ)	Key Points
Stephen 9 yrs.	11). DCD plus Dyslexia.	M-ABC 9 yrs. Total 2 VMI 9 yrs 23 VMI Non-motor 86	VIQ 126 Ed. Psych. reported sequencing problem	Presenting pain symptoms led to orthopaedic opinion Family history dyslexia, clumsiness – accepted 'clumsy' as normal.
Elizabeth 10 yrs	12). DCD and/or AS?	M-ABC 10 yrs. Total 1. Poor ball skills and balance. VMI 14 VMI Non-motor 75	VIQ 147 PIQ unstable Coding and block low	Language and social integration Exclude on Criterion C Need for screen in non-motor domains: Autism Spectrum Screen (ASSQ) abnormal.

8.1 DCD - does it exist as a syndrome?

There is no doubt that children of average or well above average intelligence whose motor development is abnormally slow do exist. Walton (1962) was one of the first clinicians to provide a fairly detailed description of these children and the first of the cases (John) is simply presented to demonstrate how history continues to repeat itself.

The relatively small number of children without any other problems has led several authors to note that pure cases are the exception rather than the rule (Hill et al., 1998; Kaplan et al., 1998). As the second case study Frank, shows, being a 'pure' case, does not mean that the difficulties experienced are any less severe.

The majority of children with 'specific' learning difficulties are boys. This applies to Dyslexia, Attention Deficit Hyperactivity Disorder (ADHD) and Asperger's Syndrome as well as DCD. No good explanation of why this is so exists. However, when girls do have DCD it is no less severe and may have an impact on the child's life that is at least equal to that experienced by boys. Annie, the third case study illustrates this point well.

8.1.1 History repeats itself: John (Case 1)

The subject of this first case study is a boy, followed over a period of several years, who bears a startling resemblance to cases reported by Walton, Gubbay and colleagues (Editorial, 1962; Gubbay, 1975). Walton highlighted the frequent finding of clumsiness present from an early age, affecting daily skills and school progress (e.g., with handwriting) yet there were minimal signs on formal neurological examination. A high percentage of the cases had reports of infections such as meningitis. The children were more often boys and left-handedness, ambidexterity or cross-laterality was common. 'Fidgetiness' was another almost universal feature. A discrepancy between Verbal and Performance IQ score on the WISC was considered a confirmatory test in identifying the 'clumsy' child.

Background and relevant early history: John's birth history was unremarkable. Although never confirmed, however a high temperature at age three weeks was queried as due to meningitis. Most children crawl before they walk but John 'bottom

shuffled' and walked at 18 months (slightly later than average). Hearing, vision and language developed normally.

Movement difficulty: signs and symptoms.

- Initial concern at 2.5 years: frequent falls and an 'odd' running style.
- Paediatric opinion "no serious abnormality" referred for physiotherapy.
- At 4.8 years physiotherapist reports continued falls. Poor gross and fine motor skill. Tense, fisted, left-handed pencil grasp.
- Sessions with private teacher for help with handwriting.

Diagnostic process: formal assessment: John was reassessed at five years by a Consultant Paediatric Neurologist who noted "some difficulty with rapid manipulative tasks with his upper limbs but no other significant neurological abnormalities" and a diagnosis of **motor dyspraxia** was made. A skin naevous (birth-mark) was noted, but investigation to rule out a neurocutaneous syndrome (such as neurofibromatosis) was declined.

John was referred to me at GOSH for assessment at age six years. His parents also arranged an independent multidisciplinary opinion (doctor, educational psychologist, occupational therapist, physiotherapist and speech and language therapist). These assessments took place at two different centres, involved many professionals, with results in agreement. John scored on the 8th percentile (borderline movement difficulty) on the M-ABC with manual dexterity, ball skills and balance sub-scores all in the borderline range. He had poor postural stability with lax ligaments and flat feet. He walked with a mild in-toe gait especially with his right leg and during assessment he once tripped on the right leg. Soft neurological signs (overflow mirror movements and facial grimace) were noted and sequencing fingers, and gesture tasks were poorly performed. Similar to the Walton (1962) case studies reported previously he was cross lateral. Subsequent re-assessment on the M-ABC at age 11 years yielded a total score of 24.5 (below 1st percentile).

Impact of the motor difficulty: John's difficulties impacted on his daily life in a variety of ways, which changed over time. Initial general gross motor difficulties were followed by difficulty learning to dress himself, handle cutlery or manage tools such as scissors. The problems singled him out as 'different' from his siblings and he

underwent many assessments by various professionals. At Primary school his class teacher noted that he was generally clumsy and that his awkward pen grasp was already compromising handwriting progress.

John was aware of his movement difficulty and commented that he wished he could avoid falling over all the time and that he hated writing. Later, his advanced reading and creative writing ability led to great frustration that handwriting was not at a similar level. In addition, the fact that his day was disrupted by attendance at physiotherapy sessions set him apart from his peers and made him feel 'different'.

Despite intervention, John's difficulties persisted and at secondary school his standardised movement and perceptual score were markedly below average. He was disappointed that he remained unable to ride a bicycle like his peers and was sad never to be selected for school sports teams. Although he typed fluently at 36 words per minute, and was progressing well academically, he preferred to use his laptop and avoided writing. His parents described his handwriting as "horrendous". However he was described as popular, well behaved, and a pleasure to have around at home.

Comment: John could easily have been among the cases which Walton labeled as "the clumsy child syndrome" (op cit.). In current DSM-IV DCD terms he would meet Criteria A, B and D but as in Walton's seminal paper, the history of infection, possibly meningitis nowadays makes John an arguable candidate for a diagnosis of really 'pure' DCD.

In contrast to what might have happened in the early sixties, John did get help from both educational and health experts. This case also demonstrates the current involvement of a great many different professionals with sometimes the danger of duplication of services. For John, modern computer technology, not available to Walton's patients, was particularly valuable.

8.1.2 'Pure' cases do exist: Frank (Case 2)

'Pure' cases of DCD appear to be in the minority but they do exist. Case study 2, Frank, who was referred at age eight years, is such an example.

Background and relevant early history: Frank was born at full term (Caesarian section) to a supportive two-parent family. Crawling, walking and language development were reported as on time and quite normal.

Motor difficulty: signs and symptoms:

- Frank's parents noticed he had more difficulty than his friends did with many everyday tasks including dressing, using cutlery and writing.
- At eight years: Frank could swim well but could not ride a bicycle.
- Frank's vocabulary and use of language was considered to be age appropriate but his articulation was rather unclear.

Diagnostic process: formal assessment: Frank's parents had been concerned about his poor motor function, but he was aged eight years before they succeeded in obtaining a referral for assessment. At that time, a consultant Paediatrician's examination revealed no hard neurological signs, no suggestion of any medical diagnosis, nor any signs of a pervasive developmental disorder. Nevertheless there was sufficient concern for Frank to be referred for a more detailed assessment of his movement difficulty.

Formal physiotherapy assessment on the M-ABC at eight years gave a total score of 15.5, i.e., below the 5th percentile. On the VMI, visual motor integration was average (39th percentile). However, the pure motor sub-test was on the 25th percentile indicating a problem in motor execution rather than pure visual perception. Observation revealed that Frank had particular difficulty organising sequenced actions such as lacing and fine motor sequences (touching each finger with his thumb, bi-directional tongue or forearm movements) were incoordinate. Writing was a struggle, spacing was largely absent, and he did not seem to be able to plan ahead but usually crossed out or corrected mistakes once they were written. Drawing a person showed that he was confused on use of space and the body parts in the figure tended to be out of alignment. Behaviour screening questionnaires (including peer relations, emotion, conduct and activity level) were in the normal range with no suggestion of problems outside the motor domain.

Two years after his initial assessment Frank's M-ABC score at age 10:11 was 11 (borderline movement problem).

Impact of the movement difficulty: According to his parents and teachers, Frank seemed to learn something one year and then 'lose it' the next which compromised progress in school. Frank complained that reading and writing were not his favourite subjects and he struggled and was anxious lest he did badly and failed. Also noted, were problems with rhythmic tasks (e.g., when the class were asked to beat time) and puzzles requiring perceptual judgements.

As Frank approached the time for transfer to secondary school, his M-ABC score had improved into the 'borderline' range. Movement problem continued to affect him in everyday life, one example being that he still preferred to use his fingers rather than cutlery to eat his food. This was not only frowned upon by his parents but also led to ridicule by his peers. At school, too, he was disappointed to fail to progress far in sport. Frank's writing problem was helped by the use of a laptop however spelling, punctuation and maths remained weak areas. His self-esteem was low as revealed by his negative comments about his errors and failures.

Comment: Frank is an example of a boy who meets all the criteria for 'pure' DCD. He is bright, with no major language problems. The M-ABC score, a reasonable estimate of where he stands in comparison with his peers, fell below the 5th percentile at age eight years, was still below the 15th percentile three years later and his VMI (visual perceptual) score placed him as low average. He showed no evidence of any medical or neurological problems, his birth history did not suggest a problem, he shows no hint of behaviour, attentional or pervasive development and he does not demonstrate associated specific learning difficulty. In spite of the 'purity' of the problem at one level, however, Frank's difficulties in school and lack of self esteem are consistent with the many reports in the literature of low self-esteem in children with DCD (Henderson, May, & Umney, 1989). Whether this is an inherent part of the child's personality, whether it follows from the functional impairment, or whether it is the interaction between the two is an open question.

8.1.3 Not all children with DCD are boys. Annie (Case 3)

In most research studies, the proportion of boys to girls is in excess of 3 to 1. In the previous study for example, there were 5-6 boys to every girl referred to GOSH. What the following case study does show, however, is that when girls do come to attention their difficulties are every bit as complex and severe as those of the boys.

Annie is a girl who was followed up over a long period at GOSH between the age of eight years to over 16 years.

Background and relevant early history: Annie was born at 37 weeks gestation by emergency LSCS following signs of foetal distress. She walked a little late but language developed on time. There was a history of clumsiness in a grand parent.

Movement difficulty: signs and symptoms.

- Parents noted that Annie, from an early age, had difficulties with most movement tasks.
- Frequent falls. Fractured wrist on two occasions.
- Before she reached school age Annie's parents searched for a reason for her problems and received a confusing assortment of labels that included 'clumsy', (which upset both Annie herself and her family), sensory integrative disorder, motor learning difficulty and developmental dyspraxia (the label her parents preferred).

Diagnostic process, formal assessment: At age eight years Annie was seen by a consultant paediatric neurologist, whose examination revealed no hard neurological signs nor evidence of any medical condition. Specifically, his report commented on the fact that reflexes were normal but mild truncal ataxia was present along with rapid manipulation difficulty. Formal assessment (M-ABC) from age eight years was consistently low and when I met Annie at age 10 years her score was 30 (i.e., below 1st percentile). On all sub-sections, she scored poorly but manual dexterity was the most impaired and her hands were noted to be a little shaky when performing precise actions such as shifting pegs or pouring a drink. At assessment Annie ran as if wearing heavy boots and she tired quickly. Visual motor integration was below normal, on the 8th percentile and by age 13 years it had improved but remained low (14th percentile).

Impact of the movement difficulty: As a small child, Annie would climb up play structures and 'freeze' unable to get down which made her frightened and led to avoiding parks and fairground rides. Although reading and comprehension were above average, writing was a problem, which affected her enthusiasm for school. In fact it was commented that even by age four years she was already reluctant to use a

pencil. She was unable to tie shoe-laces or her tie and needed help wiping after the toilet, with bathing and with washing her hair. This made Annie feel dependent and useless and she soon lacked any confidence. Transfer to secondary education was a problem for Annie as she found that carrying her school bags all day to each different classroom was both tiring and she often lost her way. Annie's balance remained somewhat insecure throughout her school life, she never learnt to ride a bicycle but she very much enjoyed horse riding. Perceptual and fine motor problems continued and she required supervision at age 16 for telling the time, kitchen safety (as she often burned herself), finding her way and negotiating crossing roads.

Comment: Annie's case is included both as an example of a girl with severe movement difficulties which persisted into adult life and also to illustrate other common perceptual problems children experience. For example, finding their way round school, anticipating traffic speed and judging where and when to cross a road safely. In addition, Annie's story also provides an example of a child who received many different labels, some of which distressed her. Various issues about use of labels is the theme in the group of case studies which now follow.

8.2 The pros and cons of labels

Debate about the use of labels in relation to many childhood disorders can become extremely heated among parents and professionals alike. Using parents as an example, one can easily find those who have been much relieved at the receipt of a label for their child's difficulties. Conversely, it is easy to find parents, who have not found it helpful at all.

The first two case studies presented next illustrate both the positive and the negative aspects of applying labels. The first boy, Kevin, provides a perfect example of the positive effects that a label can have. As a secondary issue, Kevin also illustrates the fact that the selection of which label to use may be academic and even two neurologists may not agree. Having a label at all is what matters! Adam, the second case in this group illustrates how a variety of different labels can be a source of confusion and hinder rather than help the path toward a diagnosis. Finally, Brian's experiences provide a good example of the effects that a very late diagnosis/label can have on a child and his family.

8.2.1 A label can be a great relief: Kevin (Case 4)

The primary reason for choosing this case study is to illustrate the positive side of receiving a label. Kevin also provides a very good example of all the issues surrounding a child who lies in the grey area between a firm diagnosis of mild cerebral palsy and DCD. His history reflects the typical picture portrayed in early descriptive studies, which include children with signs and symptoms suggestive of mild neurological damage or cerebral palsy (Illingworth, 1963; Gubbay; 1975).

Background and early history: Kevin was the smaller of non-identical twin brothers born at 34 weeks gestation by Caesarian section. He was born second but no hard signs of cerebral palsy were documented and his parents believed both boys to be quite normal. However the twins, although they were equally active and alert, and language was on time, developed rather differently.

Movement difficulty: signs and symptoms.

- Disparities in Kevin's and his brother's development emerged and became more pronounced over time.
- Kevin never crawled and unlike his brother tended to tip-toe walk from the beginning but his mother was told that he would grow out of this.
- Kevin learnt to ride a bicycle long after his brother and in an odd way.
- Although he could swim, he was observed to use an odd right hand motion with a screw kick.
- His mother noticed that his articulation was not always clear and he dribbled.
- Although he enjoyed jigsaws Kevin always had problems with construction kits.

By the age of nine years his method of walking was such that he wore his shoes out rapidly resulting either in expensive repairs or outlay on new shoes. At this time, Kevin's GP referred him to a physiotherapist for foot exercises. There appeared to be slight tightness in the lower leg and a paediatrician attempted to stretch the calf muscles in below knee plaster casts. This intervention produced feet flat on the floor but his parents remembered that Kevin was initially unable to walk after coming out of plasters and his gait seemed even more impaired.

Diagnostic process, formal assessment: Medically, Kevin was a fit boy with few reported illnesses. He was very short sighted in one eye and had had grommets in

both ears at school entry but hearing was reported normal. He attended the same mainstream school as his brother but Kevin's parent's concern increased as they could see that his motor skills were not at all as they should be. They began to wonder whether he might have inherited the same muscle problem that had affected a relation and finally they managed to have him referred for formal investigation at the neurology clinic in GOSH when he was 11 years old. Detailed neurological and biochemical examination revealed no neuromuscular disorder and Kevin was thought "more likely to be dyspraxic than anything else" and referred to me for physiotherapy assessment and advice. Although signs and symptoms were suggestive of underlying neurological impairment paediatric and orthopaedic consultations had never suggested that he had CP but used the label dyspraxia.

Formal assessment (M-ABC) gave a score of 24.5 (below 1st percentile) at age 11 years with difficulty evenly spread across all sub-sections of the test. On the VMI, he scored below average with a motor free visual perceptual test equally low. He was noticed sometimes to tackle geometric forms from right to left (in reverse direction). My clinical examination showed the following observations: muscle power was normal but his ankle range was reduced and the hamstring (back of leg) muscles were mildly tight and tense (hypertonic or spastic) with reduced popliteal angles especially on the left. Skipping was neither fluent nor automatic and hopping on the left leg was more difficult than on the right indicating asymmetry. Fine motor control was awkward and the quality was similar to motor patterns observed in children known to have suffered slight neurological damage. He found it hard to perform fast finger sequences and although simple articulation was clear he found imitation of mouth and tongue postures challenging. He had no problem with gesture, pantomime actions or copying rhythms. Kevin adopted a tense right dynamic tripod grasp. Letter formation was correct but spacing between words was rather inconsistent. When drawing or writing he tended to correct his work as he progressed, relying on visual feedback rather than forward planning. In contrast, qualitative observation showed that Kevin was well focussed and his effort was commendable. There was no sign of impulsivity nor hyperactivity. At this time I explained to Kevin that he presented with the sort of difficulties that might have arisen during early development and birth and that this might be termed a developmental coordination disorder or DCD.

Kevin returned for assessment at 15 years as the Special Educational Needs Coordinator (SENCO) at school felt that he should be allowed extra time in exams on account of his handwriting difficulty. Observation and timed handwriting at this time revealed neat and clear print rather than cursive style at a rate of 124 letters/minute (31 words). Even after a short period of two minutes writing, however, Kevin tended to tire and show muscle tension around the arm and neck. It was recommended that Kevin be given additional time in forthcoming GCSE examinations. He subsequently achieved excellent results and expressed his gratitude that extra time had helped him complete his GCSE exam papers.

Impact of the movement difficulty: Kevin was clumsy using cutlery and found food textures a problem which made meal times slow and messy. In sport his stiff legs, odd swimming style and unusual posture when cycling raised derisive comments from peers and his brother which made him constantly concerned that he was 'different'. In school his teachers told Kevin that he was lazy and was not making enough effort. Although Kevin voiced his main concern that his handwriting was messy and a bit slow which meant he was unsatisfied with his work, he knew that he tried hard but was frustrated and felt the teacher comments were unjust. He was losing confidence in school and felt rather depressed that he was so much less dexterous than his twin brother.

The impact of assessment, explanation and a provision of a 'label' was a positive move forward. Kevin gained confidence and following assessment said that he realised that he had probably always had a movement problem that was not his fault and was not due to lack of effort on his part. His teachers also began to understand his difficulties and the PE department, especially, began to incorporate muscle stretches appropriate for the whole class and were quick to encourage Kevin in all sporting activities. He blossomed and took on a variety of sports including fencing, water polo, and began to use touch typing for much of his school work.

Comment: Kevin is a boy who demonstrates very clearly the positive side of obtaining a label. Although apparently just as bright academically as his brother, Kevin had to live with the knowledge that he was much less well-coordinated for many years without any explanation. Although numerous professionals observed his difficulties it was not until the age of 11 that "an answer" was provided. Although he

will always demonstrate some lack of co-ordination, an explanation for his difficulties and a label allowed Kevin (his family and teachers) to better understand his symptoms. This stopped him being labeled as lazy, built his confidence and enabled him to focus on areas that could be improved and motivated him to develop alternative strategies to cope with particular problems.

In addition to the labeling issue, Kevin's story also provides a real life example of how difficult it can be to draw a line between DCD and CP. The neurological signs he showed were characteristic of a child with mild CP and should have been fairly obvious to any experienced health professional who saw him but it is doubtful whether these would be identified outside the medical realm. In the 1970s it was considered appropriate medical practice to refrain from recording medical diagnoses in educational records. In many instances a teacher would be informed that a child with, for example, severe hemiplegia was a child with 'one side of the body not working well' and no other explanation or guidance was given. Even today, some members of the medical profession may choose not to tag a child as 'mild CP' simply because such a label can carry a stigma (Goffman, 1963). One of the difficulties with this strategy, however, is that in order to appreciate the impact of a motor problem (Criterion B) a teacher needs to understand and acknowledge the probable cause of the motor difficulty. For practical purposes re the type of intervention that can help Kevin, whether he is labeled as DCD or mild CP may not be important, however his label may determine access and priority for help.

Finally, Kevin's' story reminds us of an issue crucial to research in the field. In a research study, health professionals might *exclude* Kevin from their cohort but professionals from education or psychology with little medical knowledge would no doubt *include* him. This illustrates the criticism raised in Chapter 2 that much published research lacks robust standardised selection criteria and valid comparison of the results therefore cannot be made.

8.2.2 The negative aspects of labelling: Adam (Case 5)

The next case study is included to illustrate that a collection of labels may be unhelpful. Even before school entry a child may have already picked up a heap of labels which are confusing for parents. Adam also shows that the route to both definitive diagnosis and appropriate intervention may be long and tortuous.

Background and early history: Adam's birth history was normal but he was slow to develop head control and walking was delayed (not achieved until 18-24 months). Adam was reported by his mother to have crawled on his belly 'like a maggot'. His language and social interaction developed on time, which contrasted with his obvious delay in acquiring motor competence.

Movement difficulty: signs and symptoms.

- His mother described Adam as very bendy, he often fell over, toppled off his chair when seated and he tired easily.
- She noticed that he had difficulty doing up even large buttons, cutting with scissors, and writing.

Diagnostic process, formal assessment: Extensive investigations of weak head control at age three months (including blood tests and a brain scan) ruled out muscular dystrophy and other pathologies. When Adam was aged five years he was observed to be rather active by a consultant paediatrician and he was given the label Attention Deficit Hyperactivity Disorder (ADHD). At about this time Adam met with an educational psychologist who labeled him as 'gifted' and 'dyspraxic'. Desperate to try to provide help for her son his mother took him for cranial massage. She was informed that NHS waiting lists would necessitate at least a two- year waiting time for a dyspraxia assessment by an occupational therapist so she self-referred privately to a Clinical Specialist Physiotherapist where the term DCD was used rather than dyspraxia.

At initial physiotherapy assessment at 5:6 years his M-ABC score was 24.5 (below the 1st percentile) with manual dexterity and balance the most affected areas. VMI scored above average suggesting that he did not have a problem with visual perceptual function. Adam, however, showed impressive hypermobility throughout his body (Beighton score 9/9) (Beighton, 1973; see Chapter 9 for more detail about the Beighton test). It was as if he were made of rubber rather than muscle and bone. He was active but his flexible body was not well controlled. I referred Adam for an opinion from a Consultant Rheumatologist who confirmed hypermobility related to an inherited connective tissue condition (Ehlers-Danlos Syndrome hypermobile type (formerly EDS III) and ruled out the more serious EDS vascular type (formerly EDS

IV). Adam's mother reported that her own hips click out and her aunt used to perform as a contortionist supporting the genetic basis for his flexibility.

Impact of the motor difficulty: Because Adam often fell over and lacked stamina his mother had to resort to continued intermittent use of a buggy even when he was nearly six years of age. This was tiring for his parents and made Adam feel like a baby. He often complained of aching limbs, which interfered with normal play activity and sleep at night. In school he had difficulty stabilising a pencil firmly for writing, which meant that he had insufficient control to write what he wanted. Following a daily progressive muscle-strengthening programme with low weight and high repetitions his mother reported that strength and postural control had improved. Adam complained that he found the exercises tedious and they made his muscles ache until he began to gain strength and function improved. His mother found it a challenge to help him persevere but she took the opportunity to do the programme with him in order to help build up her own muscles.

He continues to find buttons, scissors, writing, balance and running difficult and it is important that his progress should be monitored over the next few years for further problems. Adam has similar functional difficulties to many children who meet all the DSM-IV criteria for DCD. There are especially implications for handwriting due to the lax grip and shoulder instability but in his case the cause may be related to an inherited disorder of connective tissue.

Comment: With regard to labels, Adam's story highlights three important points: (i) in the absence of an evidence-based diagnosis a child may be tagged with many labels. This can have a very negative effect on both the child and his family, (ii) arriving at the right medical specialty may not be automatic, and (iii) waiting lists for therapy assessment, especially OT are unacceptably long.

As with other children in this series of case studies, Adam certainly meets DSM Criteria A and B for DCD but what about C? Many children with EDS or 'pathological' flexibility are not identified as having a medical condition and/or may never find their way to an expert who could identify the syndrome. In Adam's case, however, once the diagnosis was made by a rheumatologist he no longer meets current DSM criteria.

Adam also raises the question of whether sub-types of DCD exist. In his case hypermobility of the body framework met criteria for a definite diagnosis of EDS (excluded by Criterion C) however some reports suggest that hypermobility may be a defining feature of one subgroup of children with DCD. Similarly, researchers indicate that children such as Kevin, mentioned previously, may demonstrate a 'CP' sub-type reflecting the heterogeneity commonly seen in children with DCD (Hadders-Algra & Gramsbergen, 2003).

8.2.3 No label can be disastrous: Brian (Case 6)

Arguments concerning the pros and cons of labels take slightly different forms within different professions. Within the medical professions, a label is more closely linked to a diagnosis within a traditional medical model. Such labels are at the core of medical education, communication and collection of epidemiolgical data. Obtaining a label usually means obtaining a diagnosis and in many instances, some form of treatment then follows. In education, the situation is rather different, meaningful labels relate to a child's academic function and class teachers are unlikely to be given any but the most obvious diagnostic labels. For example, they may be told that a child is visually impaired but not that he has retinitis pigmentosa (a deteriorating condition), or that he has difficulty walking with a normal heel toe action but not that he has mild diplegic cerebral palsy. Teachers know to look out for failure to meet educational targets, to express concern about a child's recurrent absence from school or even their possession of threatening knives or similar weapons. However they do not know about the 'red flags' that might alert them to quite serious medical problems. In the case that follows there was uncertainty about the significance of a boy's apparent 'clumsiness' over a long period before it was identified as a medical problem. Confusion with a diagnosis of DCD may occur with many neuromuscular conditions.

Background and relevant early history: Brian was a nine-year old boy in a mainstream school which I visited regularly to provide physiotherapy for several children. One day a teacher in the school asked whether I would give an informal verbal opinion and advise her as to whether I thought Brian, a child whom I had never met, displayed 'clumsy' symptoms that were sufficiently serious to prompt the school to seek a referral for a medical opinion. The teacher told me Brian was in the playground with his classmates where I would be able to see him from the window.

Movement difficulty: signs and symptoms.

- Although Brian's parents had always been aware that he was rather clumsy and could not hop or skip like other children, because he otherwise seemed normal, they were only mildly concerned.
- Teachers noticed that he was poor in PE and became concerned when he presented difficulty climbing stairs and became more obviously clumsy and a safety risk.

Diagnostic process, formal assessment: Brian's route to diagnosis was serendipitous. As a favour, I agreed to observe Brian informally in the playground during his break-time. One glance at Brian's weak stance and arched back, his difficulty rising from lying on the ground, his pseudo enlarged calf muscles made me suspect serious muscular dystrophy (DMD). I suggested that the teacher was right to have concerns and that she should discuss her observations with his family. Brian was then referred for medical examination of his clumsiness where DMD was confirmed. Brian had been seen briefly by doctors in the past as he had never been able to hop but surprisingly no one raised any real concern. When Brian finally arrived for specialist medical examination including diagnostic blood tests these showed very high creatinine kinase levels indicating degeneration of muscle cells (well-advanced Duchenne muscular dystrophy DMD).

Impact of the motor difficulty: Brian was not able to join in the jumping games that his friends enjoyed. He had moved school several times on account of bullying which was usually related to his lack of motor skill and he was vulnerable in the playground. Yet the impact and significance of his signs to the family and especially to doctors who saw Brian early on, was out of proportion to the seriousness of the diagnosis. In fact following diagnosis of DMD, Brian was given a Statement of Special Educational Need, transferred into a Special School for children with physical handicaps and sadly he subsequently lost the ability to walk and became wheel-chair-bound although he remained academically able.

Comment: This lad had been unable to hop, had moved school due to being teased and bullied and had even been through school and clinic medical examinations! In the eyes of many people he was just a rather clumsy lad and it was not until the age of nine years that the true diagnosis was made. Brian must have been concerned

himself at his increasing difficulty and being finally given a cause may be seen as having a positive aspect to it. However the diagnosis of a deteriorating condition such as DMD is devastating for the family. One might consider that the delayed diagnosis gave the family a period of fortuitous unawareness of the future but on the other hand early diagnosis of an inherited condition allows for a more gradual period of adjustment and opportunity to discuss important genetic issues related to further pregnancies.

Traditionally sparse medical information was shared with education and teachers are still relatively unaware of the signs and symptoms of movement disorders. Although muscular dystrophy is very rare it should be a condition at the back of every health or educational professional's mind when an otherwise normal boy over the age of five years is unable to jump or hop.

8.3 The application of DSM-IV Criteria, A and C

The next group of four case studies all illustrate diagnostic issues related to DSM Criteria. The first case focuses on Criterion A, the following three illustrate the confusing presentation that may arise in the early stages of serious neurological and muscle conditions and show how difficult it is sometimes to apply Criterion C.

Criterion A: What about handwriting?

The research version of DSM provides us with one way of operationalising Criterion A i.e., a child must obtain a score more than 1SD below the mean on a standardised test of motor performance. As noted, elsewhere, however, the questions that then arise are: Which test? What should such a test include? How broad should it be? and so on. At the moment, the M-ABC is one test that is very commonly used as a measure of Criterion A, and most children in this series do fail this test. However, the authors, as well as the clinicians who use the test, would claim that there are children who pass the M-ABC but have real difficulties in everyday life (see Chapter 1). One of the most common components of this 'unidentified' difficulty is a problem with handwriting. Since handwriting is a complex task, involving much more than a motor component, this raises the question: Does handwriting on its own amount to a motor problem sufficient to meet Criterion A, and if so, how should we measure it?

8.3.1 How does handwriting fit into the broader picture? Gerald (Case 7)

A handwriting problem can have many causes. There are some children for whom it is simply part of a global delay encompassing language and reading problems. There are children whose speech and language is fine but reading and spelling are disproportionately slow to develop yet in every other respect they are level with or ahead of their peers. The child chosen for the next case study presented particular difficulty forming letters and reading yet he had normal hand and individual finger movements.

Background and relevant early history: Gerald was born at term and walked at one year following a period of crawling. Acquisition of language was recounted as slightly delayed but did not require intervention. Hearing tests were normal and his vision was corrected with spectacles. He was referred for physiotherapy assessment by his GP who suggested that in view of his slow writing and a reported 25-point discrepancy between verbal and performance IQ that this might indicate dyspraxia.

Movement difficulty: signs and symptoms.

- Gerald's parents and grandparents were all concerned that at age seven years he was virtually unable to write and that he appeared to be having a struggle to progress with reading.
- Gerald did not present with a general movement problem and he balanced and climbed proficiently. He enjoyed sport and was a keen footballer.

Diagnostic process, formal assessment: GP examination did not identify any medical condition other than asthma, which he had had since an infant. He was medicated for this and was otherwise fit and well and able to take part in a wide variety of sports. Formal physiotherapy assessment at age seven years gave a M-ABC score of 1.5 (in the normal range). VMI and a non-motor visual perception test were both normal (61^{st} percentile). Although Gerald passed standardised motor and perceptual tests it was the supplementary clinical observations of the quality of his performance that gave an indication of where his difficulty lay. Neurodevelopmental tests such as rapid forearm rotation (diadokokinesis), imitation of finger sequences highlighted a lack of fluency. All eye movements were jerky and he lost fixation and tended to overshoot. When sighting through a tube with each eye he always changed the hand holding the tube rather than being able to cross the midline of his body smoothly. He

was unsure when asked to show right or left hands and he was observed to change hands mid-task very frequently e.g., when completing the M-ABC lacing task. During the VMI and when drawing he revealed confusion on direction. As his parents had correctly observed, handwriting was the area that caused Gerald most difficulty. He was able to write his own name in cursive style as his family had practised this repeatedly with him. However when he attempted to form letters or numbers spontaneously, 50% were reversed. He was quite confused and was unable to match letters with sounds.

Impact of the motor difficulty: Gerald was confused and disheartened every day in school by the task of writing. Although he was of average intelligence and a keen sportsman his specific problem with handwriting was difficult for him or his family to understand. Gerald received a short intervention physiotherapy programme which included encouraging recognition of the value of his success in sport (to build confidence and peer approval) and specific handwriting-related activities. Subsequently, dyslexia was formally identified and recognised by his school and he was provided with specific educational support.

Comment: Gerald's case is a good example of the impact of an isolated movement problem affecting the normal acquisition of handwriting. As Gerald shows, this may leave a child facing each school day with understandable dread. It was Gerald's parents, rather than his teacher, who recognised that he was having real difficulty. It was they who instigated the assessments that lead to him accessing help in school. Some children with dyslexia also have other developmental conditions such as DCD but in Gerald's case the dyslexia appeared to be an isolated feature. Tests of motor competence such as the M-ABC (Henderson & Sugden, 1992), BOTMP (Bruininks, 1978), or Test of Gross Motor Function (TGMF2; Ulrich, 2000) do not include items that may identify sequencing and planning problems or overlook slight but significant disparity between function on each side of the body as scores from right and left sides may be combined. The supplementary qualitative observations (e.g., in the M-ABC) and neurodevelopmental clinical screening routines are particularly helpful in teasing out some of the more subtle aspects of motor performance.

8.4 The application of DSM-IV Criterion C: signs and symptoms of medical conditions

In Chapter 2 many medical conditions were listed with signs and symptoms of 'clumsiness' confusingly similar in some respects to those seen in children with DCD. Three cases are presented in the following section to illustrate and stress that teachers must be aware of 'red flag' symptoms as a child's symptoms of 'clumsiness' may be those not of DCD but of a quite separate medical condition.

The first case in this section is a boy chosen to illustrate how symptoms specific to a medical condition had a very typical diurnal pattern. Two different medical conditions are then presented which show further problems in the application of Criterion C especially when there is a longstanding history of 'clumsiness' or a condition that is only fairly recently becoming more recognised.

8.4.1 Ruling out childhood arthritis: Peter (Case 8)

This case study features a boy who presented with possible DCD and was referred several years ago to our community physiotherapy clinic. One of the key features, here, that would rule out DCD was variation of symptoms during the day, not something that his teachers had really picked up.

Background and relevant early history: Peter was a seven year old boy who was born at full term by normal uneventful delivery. His milestones were mildly delayed.

Movement difficulty: signs and symptoms.

- Peter was noticed by his parents and his teacher to fall more often than his class-mates and to seem generally clumsy and awkward.

Diagnostic process, formal assessment: Peter was referred to the community paediatric service for initial physiotherapy opinion regarding his apparent clumsiness. At this time, no standardised test was available to measure the degree of movement difficulty compared to age peers however observation of his physical activity supported his parent's concern. Careful physiotherapy examination showed slightly reduced range of movement and mild tenderness and swelling around one or two joints. Questioning him about joint tenderness revealed that there was a definite

pattern of more aches and pain first thing in the morning. Although his movement competence appeared not to be age appropriate this was suspected to be due to early Juvenile Idiopathic Arthritis (JIA) which features significant early morning joint stiffness. This diagnosis was later confirmed by the community paediatrician, rheumatology examination and blood tests. The presence of a medical condition such as JIA excludes a diagnosis of DCD according to DSM-IV criteria.

Impact of the movement difficulty: Peter had difficulty in PE and falls, complaints of aches and pains and lack of manual dexterity were at times interfering with his ability to join in normal school activity. He avoided rough and tumble play with friends at break times in order to protect his joints from bumps and strains.

Comment: Although Peter meets criteria A, and B for a diagnosis of DCD, specific signs and clues on clinical examination alerted the physiotherapist to a quite different cause for his movement problem and clumsiness. It is possible that Peter might have been 'clumsy' before he had any active arthritis. Alternatively, the juvenile form of arthritis was entirely the cause of his lack of motor competence. Whatever the outcome of this debate, Peter's case underlines the importance of ensuring that a health professional be involved in the assessment of children with movement difficulties. The treatment of children with JIA is quite different to the intervention indicated to help children with DCD. In the former medication, splinting, correct positioning and a daily exercise schedule are indicated together perhaps with adapted tools and aids for mobility. Note also that Peter's symptoms varied during the day, which may lead to significant signs being overlooked. In certain conditions problems may be more obvious when a child is tired e.g., at the end of a day or following strenuous exercise (cardiac problems) but in Peter's case it was the symptom of more aches and pain at the start of the day that supported his eventual diagnosis.

8.4.2 Chicken and egg - brain tumour or DCD - which came first? Michael (Case 9)

A brain tumour is fortunately an unusually rare condition and in the case of Michael it was quite benign. Michael is included in this series because his early history is indistinguishable from a child with DCD and begs the question: which came first the signs of a tumour or symptoms of long standing DCD?

Background and relevant early history: Michael was born at full term by normal delivery. He walked late at 22 months following a period of crawling. Language development was reported as delayed. He also had grommits inserted for a conductive hearing loss and he attended an optometrist for eye exercises when young to help with convergence.

Movement difficulty: signs and symptoms.

- Michael's parents reported that he had always been rather clumsy, accident-prone and had had problems learning to skip, using scissors, dressing, holding his pencil, learning to write, beating time.
- He was ambidextrous for many activities but used his left hand to write.

Diagnostic process, formal assessment: Localised neurological signs led to the diagnosis and removal of a benign frontal brain tumour when Michael was aged nine years. Two years later at age 11years, when he was first referred for physiotherapy evaluation, his M-ABC score was 21 (below 1st percentile). He achieved a full score on the ball skills sub-test but on the manual dexterity and balance components he scored poorly. On the VMI, he scored on the 47th percentile and a non-motor visual perceptual test was above average. At this time an educational psychology assessment reported a 45-point discrepancy between his verbal and performance IQ. My clinical observation showed that Michael, similar to Kevin, had some lack of leg flexibility with reduced popliteal angles. Although left handed, he sighted with his right eye and like many of the cases reported earlier he demonstrated difficulty in bilateral actions, direction and crossing midline. Interestingly his mother reported that she had always had difficulty on direction and she has to turn a map around to work out right/left/east/west.

Impact of the motor difficulty: Michael had had difficulty learning to write which compromised school progress. At age 10, he still could not master shoelaces, which annoyed him. However, this was a longstanding problem and not something that arose following the neurosurgery. He was keen to succeed and became frustrated and anxious when simple every day tasks presented such a challenge. Michael concentrated well and with determination he made good progress with improved flexibility and power but manipulating small objects e.g., pegs remained a challenge. By acquiring proficient key-boarding skills (50 words/minute) Michael adapted to

and largely overcame his residual motor problem however he comments that a lap-top is heavy to carry round school all day. Despite clumsiness he enjoyed and was competent at a very wide range of sports.

Comment: Michael meets Criteria A and B with handwriting once again being an area of motor difficulty which made a big impact on his progress in school. If one examines Michael's case history in the context of Criterion C, he is excluded because of a neurological diagnosis. One might presume that Michael's difficulties were associated with the neurological diagnosis since a cerebral tumour can grow slowly and may be present for some years before localising signs appear. However Michael's very early history is quite typical of children with DCD and it remains possible that part of his clumsiness was unrelated to the later pathology. One may argue that he has DCD as a discreet syndrome, that his 'Clumsiness' is symptomatic of the effect of the tumour or that DCD and the tumour are two conditions which in his case are co-ocurring either associated or non-associated. This case also highlights the different motor profiles revealed by the M-ABC and for Michael ball skills were unaffected.

8.4.3 NF1 an under-diagnosed condition: Mary (Case 10)

The last case in this section, Mary, provides an example of another condition which may be confused with DCD, Neurofibromatosis Type 1 (NF1). This genetic condition, which is reported to be under-diagnosed may account for some children who receive the label DCD. Two children in addition to Mary, both referred to me with 'DCD', were later diagnosed with NF1. NF1 features fibroma or lumps, which form in any part of the neurological system and interfere with function. Similar to Michael's case it is important to be on the lookout for localising neurological signs which would point to pathology 'red flags'. Mary again may be a clumsy child with NF1 but her symptoms are more likely to be due to the NF1 growths.

Background and relevant history: Mary was born at full term by normal delivery. She walked at 16 months following a normal period of crawling and language developed on time. Mary had a long history of delayed motor development and like cases in the first section, had received many labels to account for her problems, including developmental delay, dyslexia, and dyspraxia.

Movement difficulty: signs and symptoms.

- Mary had been labeled as a bit clumsy and with coordination problems since childhood and had attended both individual and group physiotherapy in the past.
- She was reported to have always lacked stamina. She was overweight, running was poor and she could neither skip nor ride a bicycle.
- Maps and jigsaw puzzles were difficult.

Diagnostic process, formal assessment: Medical examination at age 10 years related to skin signs led to investigations. These revealed that Mary had several small neurofibroma in different areas of her body which were compromising vision and movement. She showed typical skin pigmentation diagnostic of neurofibromatosis type 1. Formal physiotherapy assessment revealed a M-ABC score of 24.5 (below the 1st percentile) with a good score for manual dexterity but balance was below the 1st percentile and ball skills were also immature. VMI scored on the 10th percentile but her non-motor visual perception sub-test scored well above average on the 88th percentile.

Impact of the motor difficulty: Mary hated sports day because she was not athletic and her motor function was not as competent as her peers. She particularly regretted that she could not ride a bicycle like her friends. Climbing down the steps from a bus was quite difficult which made independent transport hard. NF1 is a hereditary disorder and at present there is no cure. Symptoms are highly variable and severity cannot be predicted so that the future for Mary is uncertain and dependent upon the progression of her present fibroma and whether and where further neurofibroma may arise.

Comment: The primary reason for including Mary in this series is that she provides a perfect example of a child who meets Criteria A and B and without medical input might end up with the label DCD. When all three criteria A, B and C are applied properly, however, Mary would be excluded on the grounds that the NF1 is a clearly understood 'medical' condition with known aetiology. Having said this, however, it is still possible that Mary was actually a clumsy girl who developed NF1 at a later date i.e., not all of her motor problems might have been due to the current pathology. What is important here, is that Mary required detailed paediatric neurological tests to reveal the diagnosis. It is also important that although NF1 is not common

(prevalence 1:4000; Huson et al., 1988) it has been identified in all ethnic groups and is reportedly under-diagnosed.

As a secondary issue, Mary's developmental history also presents two other points. First, Mary illustrates the fact that even children whose clumsiness does have a known medical cause can receive a collection of other labels as they go along. Second, she illustrates once again, the differing motor profiles that can be obtained on the M-ABC sub-tests. The fact that her balance was much poorer than manual dexterity may be due to her NF1 but balance may be further compromised by her high body mass index.

8.5 The application of DSM-IV Criteria: Dealing with co-occurring diagnoses

In the previous section case studies have mainly related to concepts raised in Chapters 1 and 2 of the thesis. The next two case studies reflect the ideas presented in chapter 3 where DCD is conceived as one of several co-occurring developmental disorders.

The first case, Stephen, rather than 'pure' dyslexia has problems with reading co-occurring with movement difficulty. The symptoms highlighted by Stephen led to an orthopaedic investigation where concern about coordination was raised. His case demonstrates that whether a child stands out as being 'clumsy' may depend on the perception of 'normal' movement skill in any given family.

The last child presented in this series of case studies, features a girl, Elizabeth, with DCD and Asperger's syndrome. Her case demonstrates issues related to capturing the whole picture or profile of a child and the importance of a broad assessment.

8.5.1 DCD plus Dyslexia. Stephen (Case 11)

Dyslexia may co-occur with DCD and unlike Gerald who had an isolated dysgraphia/dyslexia with a normal M-ABC score, the present case, Stephen, is included to show how significant motor problems can co-occur with dyslexia. The case is included also, to illustrate how recognition of a child's symptoms may depend to some extent on the impact or perception of 'clumsiness' made within the family. This case also highlights that initial presentation may be only part, the tip of the

iceberg, of a more complex picture. The symptom that leads to a medical examination may point to and open the doorway to investigation of other undetected signs of co-ocurring conditions.

Background and relevant early history: Stephen was born at term by normal delivery and developmental milestones were on time. He was initially referred at age nine years to a Consultant Orthopaedic Surgeon to investigate the cause of tight muscles and persistent pain in the lower legs (shin pain).

Movement difficulty: signs and symptoms.

- Stephen's motor function was not noted to be especially different to the rest of his family but pain in his legs rather than clumsiness led to investigation.
- His parents said he was rather clumsy handed and his teachers had remarked on his awkward pen grip.

Diagnostic process, formal assessment: The initial orthopaedic assessment revealed no major pathology other than the hamstring muscle tightness, which was not associated with increased tone or neurological signs. However as the orthopaedic physiotherapist was concerned at signs of a more general lack of co-ordination, after discussion with the orthopaedic consultant and Stephen's parents, he was referred for further physiotherapy evaluation using standardised movement and perceptual tests and assessment of writing and behaviour. When I assessed Stephen at age 9:6 his M-ABC score was 17 (below the 5th percentile) indicating a movement difficulty compared to age peers. He showed weak manual dexterity and ball skills and unstable static balance but comparatively good dynamic balance. There appeared to be difficulty in planning and sequencing actions. VMI was low average, on the 23rd percentile, whereas pure visual perception (without the motor component) was above average. Writing observation showed a firm right handed pen grasp and his slouched posture reflected his high flexibility Beighton score (> 6/9) and generally low muscle tone: he propped his trunk against the desk, bent over and held his head near to his work with his mouth loosely open. Discussion with Stephen's parents revealed that he had previously attended a dyslexia centre and was receiving help from a dyslexia specialist. There was a family history of dyslexia, clumsiness, flexible (bendy) joints and immune disorders. Behaviour screening questionnaires were normal suggesting no associated emotional, behavioural or pervasive developmental disorder.

Impact of the motor difficulty: Stephen himself commented: '*writing is not my best area*'. Writing was legible when he wrote slowly, but when he tried to write faster speed, accuracy and spacing deteriorated. At school, teachers described him as verbally fast but his output was slow with sequencing problems affecting progress in reading and spelling. There were no comments about lack of co-operation but he showed evidence of beginning to lack confidence and feared failure.

Comment: Stephen illustrates that DCD may co-occur with another developmental disorder such as dyslexia but without any associated emotional or behavioural problems. He meets Criteria A, B and C as he has no neurological or muscular cause for the difficulty. Stephen was identified as having a specific learning difficulty (dyslexia) but perhaps because clumsiness was apparent in relatives his lack of age appropriate motor competence was not recognised nor perceived as abnormal. Attention was drawn to the presence of the motor problem (DCD) by what appeared to be an unrelated symptom, pain in his legs, which led him through the orthopaedic doorway toward further assessment.

DSM-IV Criterion C excludes the presence of pervasive developmental disorders e.g., autistic continuum. Many children have been referred for physiotherapy assessment with movement difficulty (possibly DCD) but who also meet criteria for Asperger's syndrome. The final case study features a girl with AS.

8.5.2 DCD and/or AS? Elizabeth (Case 12)

Elizabeth was a girl who at age 10 years was thought to show possible signs of Asperger's Syndrome. Prior to full psychiatric assessment she was referred for physiotherapy evaluation of her fine and gross perceptual motor function.

Background and relevant history: Elizabeth was born at term by normal delivery and walked at 14 months but never crawled. She had mild hearing difficulty (glue ears) which were treated by the insertion of grommets and she was followed up by an otolaryngologist for several years. Her language developed on time.

Movement difficulty: signs and symptoms:

Elizabeth's parents noticed that she developed a little differently to her friends. She was always rather difficult and fussy. She hated hair washing, the texture of certain

materials and foods and loathed the feel of high-necked sweaters. She feared heights and avoided walking over any wobbly surfaces.
She was never good at sport. They felt she was disorganised and muddled when tackling any task.

Diagnostic process, formal assessment: M-ABC at 10 years was below the 1st percentile (total score 20.5) with almost full score for manual dexterity but very weak ball skills and balance. VMI was below average (14th percentile) contrasting with pure visual perception on the 79th percentile. Flexibility was average. Elizabeth demonstrated some difficulties planning actions and with direction however her writing and drawing were neat. Elizabeth was observed to have difficulty modulating her response to sensation especially touch and there were concerns regarding social integration and language processing. Her score on a short screening questionnaire for high functioning autism spectrum disorder (Ehlers et al., 1999) suggested that her perceptual motor difficulties might be a symptom of a primary problem outside the motor domain. Subsequently, a diagnosis of Asperger's Syndrome (AS) was made by a consultant psychiatrist on formal tests and in depth interview.

Impact of the motor difficult: Elizabeth had no interest in sport and the PE curriculum in school was tedious and not her preferred subject. She found buttons, ties and laces hard which made getting ready for school a chore each morning. She became obsessively interested in one topic, that would absorb her and tended to isolate her from her peers and she found it hard to make friendships. Her preference for certain sensations and avoidance of others compromised comfort and restricted her environments.

Comment: If Criterion C is strictly adhered to Elizabeth cannot be diagnosed as both AS and DCD. DCD excludes AS, however AS includes symptoms of clumsiness. This case features an uneven motor profile with good dexterity but poor ball skills and balance items. There is disagreement on which category a child such as Elizabeth should be placed. Without an appropriate screening questionnaire Elizabeth may have been viewed as a slightly odd girl with DCD. It illustrates the importance of incorporating some form of screening to identify children with possible autistic spectrum disorders.

8.6 Summary

The 12 case studies presented in this chapter bring together the real life experiences of children with movement difficulties, formal test results and observations from teachers and clinicians. As case histories, it is hoped that they speak for themselves by illustrating the many difficulties that children with DCD, and their families, encounter, thus supplementing the group data presented in the previous two studies.

Beginning with theoretical issues, the data provided by the case studies focus attention on some of the problems still to be solved if a diagnostic system based on something like DSM-IV or ICD-10 is to be made to work. Although the APA proposals are used as examples, the criticisms levelled at DSM apply equally well to the ICD-10 manual. As noted in chapter one, the criteria suggested in each manual may drive the diagnostic process in many centres, but operationalisation and interpretation is not straightforward whatever the condition being discussed. At this point, too, it might be useful to make clear the gulf between clinicians and researchers in the UK and their familiarity with these manuals. Study 1 (Chapter 5) showed that a third of professionals asked to define DCD were unable to do so and clearly had little knowledge of either DSM-IV or ICD-10. In paediatric clinical practice, especially in the community where case loads are large and varied, the majority of therapists have never heard of DSM and they base their 'diagnosis' of DCD on clinical judgement of 'clumsiness' and possibly but not necessarily, a standardised measure such as the M-ABC or BOTMP. In the commentary that follows, therefore, the reader should keep in mind that the writer is not only a therapist, working in a specialist hospital, but is also involved in the development of the paediatric physiotherapy profession as a researcher.

Against this background, the four DSM criteria are now re-considered as they currently exist. All 12 children had been referred to the GOSH physiotherapy department because of concern for some aspect of their motor performance. They were, therefore, a good sample on which to test one way of operationalising criterion A. Although there is no universally accepted gold standard for operationalising criterion A, both of the tests used in this project, the M-ABC and VMI, are recognised internationally as useful instruments in this process. Taken together, these two tests identified all but one child, Gerald, in this series of case studies as having a significant motor problem.

The fact that most children in this series - and their parents - were able to voice their concern about the way in which the movement problem affected their daily lives suggested that criterion B is relatively easy to deal with. However, there are times when the judgement about impact is not so easy. By definition, the question of whether a child meets Criterion B is based on clinical judgement - but whose clinical judgement? One particular case in this series highlights the fact that the impact of the motor difficulty may not appear the same to all concerned. In Stephen's case, his M-ABC score fell below the 5th percentile and all of the professionals involved perceived a problem but his parents did **not.** They seemed to see his motor function as no different to the rest of the family. Conversely, other children, not included in the cases, arrive at physiotherapy assessment because of parents' concern at lack of neat writing (sometimes as young as age 4 years!) yet the professional involved sees no sign of a movement problem. Instead what they perceive is a child pressured by demanding parents and a high achieving academic school. Who may be right in the long term, however, remains an open question.

That children's movement difficulty is observed to change over time brings with it theoretical questions of whether a child can move into and out of a DCD category? Some children such as Kevin in this series improved or coped better over time but others continued to have problems (Annie). A child may just cope with handwriting lessons, and with ball games with a static partner, during Primary school. The same child may be unable to adapt at the Secondary stage where there is greater demand to write at speed or join in complex ball games where both the child and team-mates around him/her are constantly moving. Theoretically, evidence of such change over time, argues for a flexible system, where labels are attached and removed as appropriate. In reality, once a label has been applied it may be difficult to dislodge.

Numerous studies report that handwriting is of major concern for children with possible DCD and is often the problem that leads to referral. The cases reported here support this claim with at least 70% experiencing a problem with handwriting. The fact that handwriting is a complex skill which is difficult to assess and often complicated by other, related literacy difficulties makes it difficult to decide whether it could or even should be part of a battery designed to fulfill Criterion A.

One of the issues rarely addressed in DSM or ICD is the question of subtypes within the conditions listed. At a functional level, the children described in these case studies showed quite different profiles on their M-ABC and VMI test results. On the M-ABC, for example, there was a range of severity within the group with some children scoring below the 1st percentile others barely below the norm. There were some children who showed only gross motor difficulty (e.g., Brian), and others where fine motor problems were of most concern (e.g., Annie). These patterns then combined with both pass and failure on the VMI (e.g., Adam and Michael score below the 5th percentile on M-ABC and passed the VMI; Annie, and Mary scored below the 10th percentile on both). On the VMI, the children also ranged from those with no difficulties (John) to others who featured very poor scores (Annie). Yet others were rather better on the purely visual aspect compared to the sub-section with a motor component (Mary). Even on these two tests alone, therefore, there was some support for the idea that specific subtypes within DCD might exist, but confirmation of this would require a quite different sort of study.

Although the application of DSM Criteria A and B raise some difficulties, there is no doubt that the application of Criterion C is the most problematic. One of the major difficulties is clearly illustrated by the children in this series who presented with 'excluded' medical diagnoses. As Mary's case illustrates, the diagnosis of many such conditions is not simple, may emerge over considerable time and depends upon access to expert medical opinion. The many practical issues this process raises is discussed further below. Another problem associated with the application of Criterion C concerns the dividing line between conditions such as CP or connective tissue disorders. Among the 12 case studies presented in this chapter, there were children who featured symptoms similar to mild CP (Kevin) or joint hypermobility (Adam). In these cases, it was not easy for the expert involved to draw a neat line between a diagnosis of DCD and the alternative conditions - and perhaps the reality is that this is not actually possible. Conditions like DCD may be inherently 'fuzzy-edged' and diagnostic systems of the future must somehow reflect these continua.

One of the most difficult issues to deal with in relation to the use of DSM relates to the way co-morbidity is dealt with. The manual directs us to exclude children who have a medical, neurological or pervasive developmental disorder. As has been discussed in Chapter 3, DCD is viewed by some people as one of a group of

developmental conditions with a common underlying cause/s in contrast to DSM which classifies and separates developmental conditions into discreet disorders. Conditions that are known to co-occur with DCD are featured in many of the case studies presented and the child such as Frank with 'pure' motor difficulty is in the minority. This emphasises the importance of a broad assessment battery to determine either the weighting of the 'motor' problem or rule out the impact of non-motor aspects. One way of thinking of DCD is related to the fact that symptoms evolve during development and the picture is not static. Is DCD a temporary 'sorting' house until the child's problem becomes more clearly defined?

Turning to more practical issues highlighted by these case studies, a rather obvious place to start concerns labels and their acquisition. Several of the children described in this series had received more than one label on their journey to a 'movement related' one. Some had had a positive impact, others a negative one. In cases where multiple labels had been assigned, a related problem, of course, was the time which had elapsed before some families reached any sort of explanation of their concerns about their child. Numbers on a page, showing that arrival at a diagnosis can take many years somehow fails to convey the anguish that parents must experience as they move from one clinic to another, seeing the waiting times lengthening and their child getting older and help as far away as ever. A comment from a referrer apropos the request for assessment stated "*This child has no chance of getting an OT assessment or help during his Primary school years, please would you assess his perceptual motor function and give these parents some suggestions*". In some of the cases reported here, there were understandable reasons for delays. Sometimes the route taken was via many different clinics and the diagnosis was by necessity an evolving process. In one case, for example, what began as DCD progressed to a later diagnosis of Asperger syndrome (Elizabeth). Conversely what started out as an orthopaedic referral (Stephen) turned out to be merely the tip of the iceberg and just one symptom which started the diagnostic process. Mary and Michael may have had long- standing DCD but other pathologies developed and became identifiable over time. In other cases, however, delays are not explicable and long waiting lists are of great concern (Dunford & Richards, 2003). This raises practical issues of the time and cost involved in multiple assessments, the question of whether intervention is evidence based and effective and how professionals and non-professionals skills are utilised in the assessment/ intervention/evaluation process.

There is another dimension to the labeling issue that has not yet been mentioned, that is the fact that a child's label can often reflect the 'expert' who carries out the assessment rather than the whole picture of the child. Many of the current case study children mentioned their disappointment in sport, inability to ride a bike, or play ball games with their friends. In some instances, GOSH physiotherapy department was their first port of call and the mention of problems in riding a bicycle etc alerted the assessor to possible balance problems, hand-eye coordination problems etc - and thence to the label DCD. However, there were other children who had seen other 'experts' first and they had not focused on the motor difficulties the children experienced. These children had received alternative labels because other difficulties the child was experiencing were salient for that particular assessor. To complicate this issue further, the literature on childhood disorders in general shows clearly that children's profiles of performance both within and across domains are not static but often change over time. In this series of case studies, the trend throughout was for **motor** problems to continue into secondary school and beyond. However, in other regards the picture was less clear. In some cases, the child seemed to cope better e.g., Kevin but in others new problems emerged as adulthood approached (Annie). This variation raises practical questions regarding when intervention is most appropriate, who should be the key person involved with the child and the importance of planning ahead for whatever action might be needed.

The confusion between DCD and medical conditions such as DMD, cerebral tumour, NF1, JIA, Ehlers Danlos and CP referred to previously raises a practical issue that affects both health and educational professionals - the question of 'red flags'. From the medical perspective the problem is how to describe such tell-tale symptoms as muscle weakness without frightening the teacher or causing every tired child to be referred. For the educationalist the concern is how to allow a child independence in school without compromising safety and ensuring that 'red flags' do not cause panic but are dealt with according to a clear action protocol.

Finally, a major problem for all professions together concerns how one might capture a picture of the child's function across all domains in the limited time available for assessment? Perceptual-motor and handwriting tests address core aspects of DCD but what of other things? In order to address the issue of DSM Criterion C children must

also undergo medical screening, which can be time consuming, especially if really puzzling symptoms emerge. Also, consideration must be given to the impact of any educational, psychosocial and/or emotional problems on the motor difficulties and vice-versa - and in conclusion, without some overview of the family dynamics a complete picture of the child as a functional being in a complex environment will never be obtained. Fortunately, some of these issues impact more on clinical practice than as a primary concern in a research project.

The final three pages of this chapter include photographs that illustrate points raised in the case studies related to clinical assessment. Included in this section are photographs that show joint hypermobility, examples from the M-ABC and VMI tests, samples of handwriting and a series of drawings. Together they capture the variey of function observed in children with DCD and other neurodevelopmental disorders and medical conditons.

Figures 8.1-8.8 *Examples of hypermobility, figure drawing, handwriting, M-ABC and VMI tests*

Figures 8.1a and b show examples of excess range of joint hypermobility in a boy aged 7 years.

Figure 8.1a

Figure 8.1b

Figures 8.2a and 8.2b illustrate two different approaches to the M-ABC 'Flower trail' item. Errors are shown in both drawings, where the trace moves outside the track, but additionally the focus of the task is rather different in Figure 8.2b.

Figure 8.2a *Aged 7yrs (HFA)*

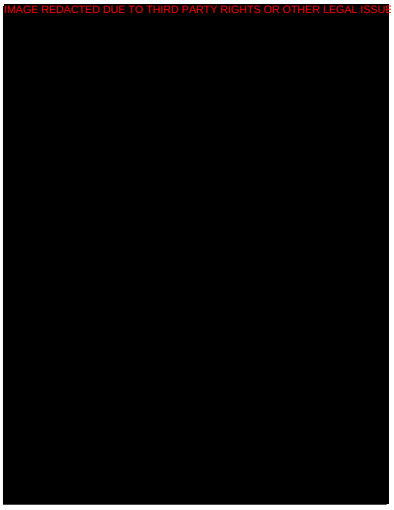


Figure 8.2b *Aged 8 yrs (DCD+)*

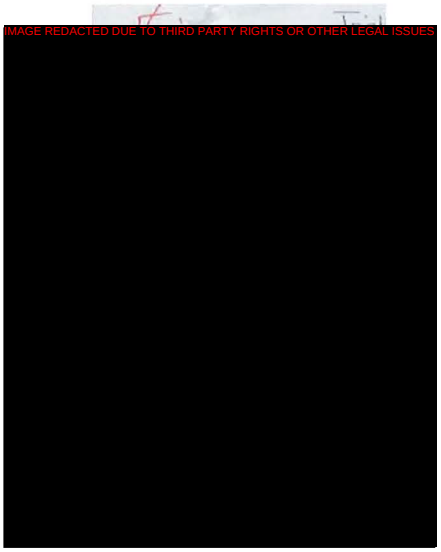


Figure 8.3 typical child aged 7 years, well organised for the start of the M-ABC Peg-placing task

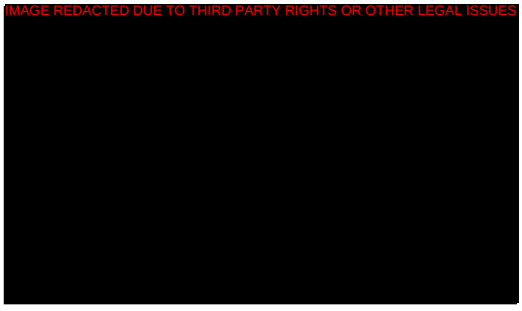
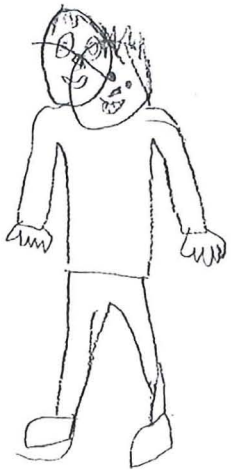


Figure 8.4a and 8.4b show visual spatial and difficulty in planning symmetrical body scheme by two children presented in the case studies a) Annie and b) Freddie.

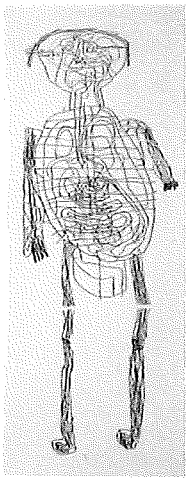
8.4a



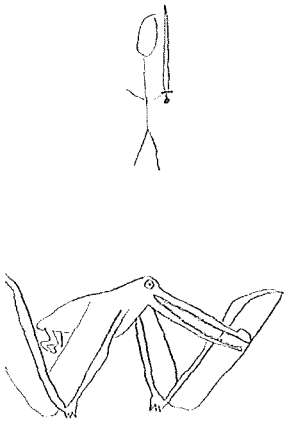
8.4b

Figures 8.5 a-c show two drawings of 'a person' by children with HFA (on the left) and by a typically developing child (on the right). The HFA drawings show unusual attention to detail but often lack human facial features.

8.5a



8.5b



8.5c

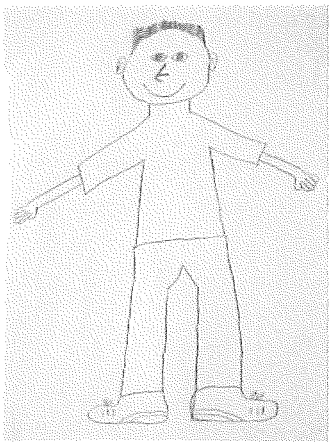


Figure 8.6a and b Writing Samples by typically developing children aged 7 and 10 years

Figure 8.6a Aged 7.8 yrs

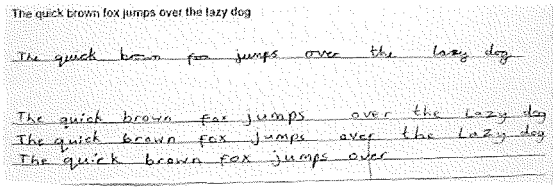
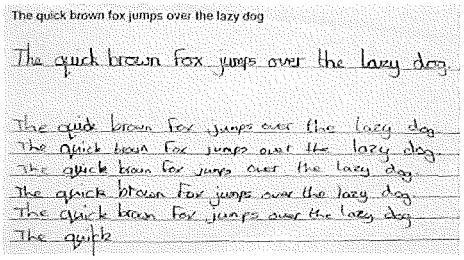
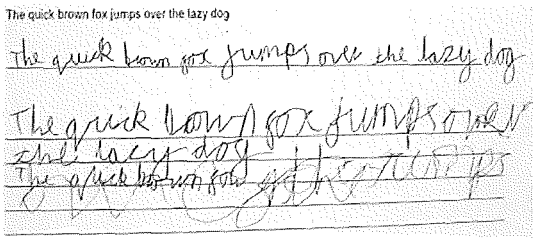


Figure 8.6b Aged 10.1yrs



Figures 8.7 a and b Writing Samples by children a) with DCD and b) with HFA/AS

8.7a Aged 7:8 (DCD)



8.7b Aged 9.7 (HFA/AS)

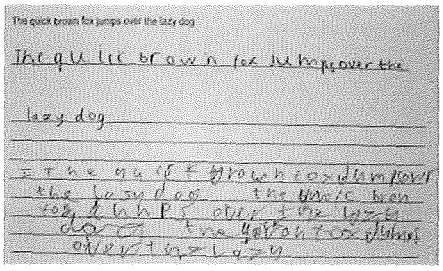
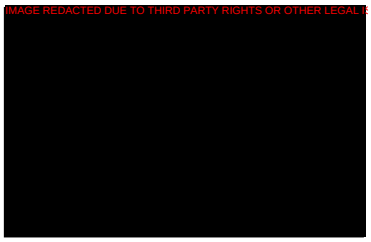


Figure 8.8 Case study 'John' showing awkward grip but legible writing at age 11:11



Figures 8.8 a-c show samples of the different aspects of the VMI. (a) Confusion on the main VMI test (b) Reversal in the visual sub-test (c) Deviation outside the marked lines on the Motor sub-test.

Figure 8.8a

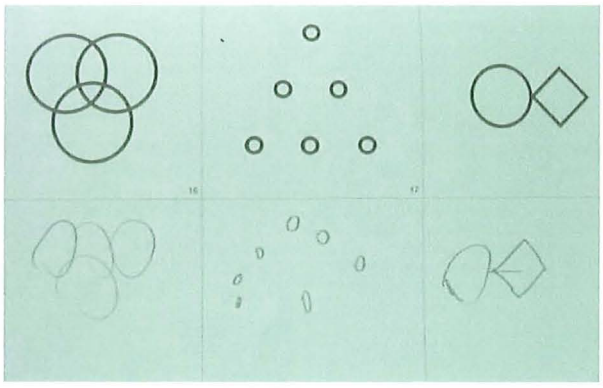


Figure 8.8b

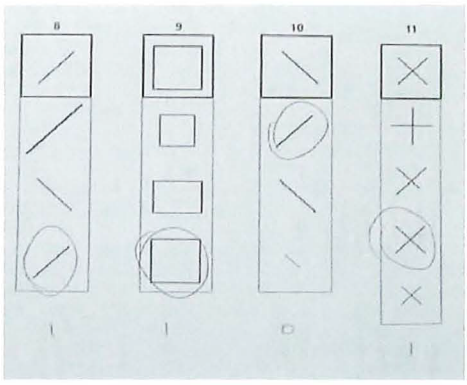
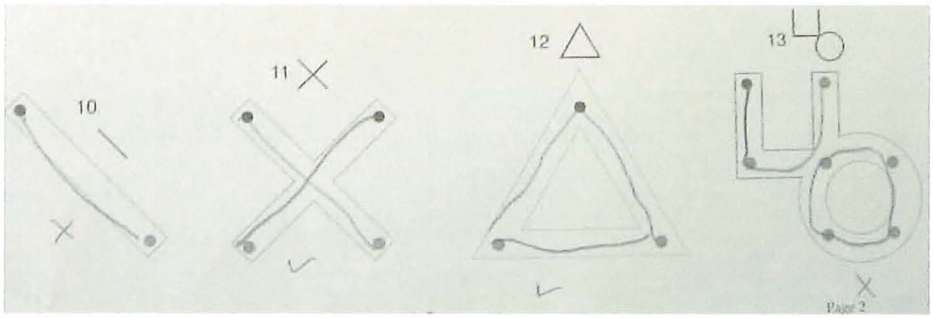


Figure 8.8c



Chapter 9

Study 5 DCD: A Separate Syndrome or Not?

9.0 Introduction

The central theme running through this thesis is the question of (a) whether DCD exists as a separate syndrome, and (b) if it exists, how it differs from other conditions which have clumsiness of movement as a feature. So far, the studies reported support the notion that DCD does exist as a separate syndrome, but do not address the second part of the question. In the present study, both of these questions are addressed directly. Planning a study designed to address such questions, raises two important design issues. The first concerns the choice of children to be included, the second concerns the kinds of tests one might employ to explore differences between the selected groups.

Starting with the choice of children, if one assumes for the moment, that DCD does exist, with whom should children with DCD be compared? As noted in Chapters 2 and 3, clumsiness of movement is a common feature of many childhood diseases and disorders. Chapter 2 discussed 'clumsy' symptoms observed in conditions, such as cerebral palsy (CP) and childhood arthritis, that might be broadly classified as having a 'physical' origin. In contrast, Chapter 3 considered syndromes also featuring 'clumsiness' but which are loosely grouped under the heading 'specific' learning difficulties and are mainly referred to educational or clinical psychologists, or psychiatrists - e.g., dyslexia, specific language impairment (SLI), AS and ADHD. Originally, the design of the present study required inclusion of two groups from each category 'physical' and 'psychological'. However, practical considerations intervened and the initial plan was somewhat reduced in scale.

From the possible set of physical conditions for example, neurofibromatosis type 1 (NF1) in which there are perceptual, motor and handwriting problems (Hyman et al., 2005) and benign joint hypermobility syndrome (BJHS) were considered seriously. However, the former, NF1, which is under-diagnosed and may well be confused with DCD, was rejected on the grounds that presentation is specifically determined by the site of the tumour/s. In contrast, the condition benign joint hypermobility syndrome (BJHS) seemed to be a better possibility and the rheumatology department in GOSH,

which runs a hypermobility clinic, was interested in collaboration. As noted earlier, the defining feature of BJHS is an excessive range of movement in joints due to ligamentous laxity, associated with musculoskeletal symptoms. The syndrome affects muscle and joint elasticity giving rise to subluxations and strains, 'growing pains', and backache. Also, Murray and Woo (2001) note 'unusual or bizarre hand postures' which cause pain and fatigue in the hand and wrist compromising handwriting. In addition, BJHS involves many tissues throughout the body and is reported to be associated with anxiety, depression, fibromyalgia and chronic fatigue (Grahame, 2003, p. 11, Box 1.5; Barron et al., 2002). Like many other syndromes, which seem easy to diagnose when the symptoms are simply listed in black and white, however, BJHS turns out to be less easy to diagnose in practice. One of the difficulties is that hypermobility, without the associated features, occurs as a 'symptom' in a significant number of apparently normal children of primary school age with a figure around 7% quoted for Caucasian boys, almost double this figure in girls (Mailard & Murry, 2003, p. 37) and is increased in various ethnic groups e.g., West Africans. What this means, therefore, is that the diagnosis of BJHS is dependent on the presence of aches and pains etc, which some parents may feel warrants further investigation and others may simply ignore in their offspring. Thus, who reports such symptoms and which children get referred to a rheumatology clinic may make the difference between 'hypermobility' as a symptom and BJHS as a syndrome indistinct. By virtue of their clinical referral, boys in the present study were presumed to meet criteria for BJHS (but see below for further discussion).

From the set of conditions, that may be loosely labelled as 'specific' learning difficulties, the possibilities initially appeared to be excellent. At GOSH, two specialist clinics within psychiatry are well established and are very research oriented. They see children with Tourette's Syndrome (TS), and with High Functioning Autism. Also, there were strong links with local community clinics where children with ADHD are followed up. Each of these conditions may feature a specific type of movement problem as well as general movement 'awkwardness': Tourette's features sudden involuntary actions (tics), Asperger's features idiosyncratic actions (hand flapping) and in ADHD there is an excess of physical activity. Unfortunately, however, neither the Tourette's nor ADHD referral sources were able to refer suitable participants, leaving only an HFA/AS group (see method section for reasons given).

Just as the diagnosis and labelling within the field of DCD has undergone discussion, argument and change over time, the same has occurred around the terms High Functioning Autism and Asperger Syndrome. Although Asperger Syndrome is currently given a specific entry separate from autism (which includes high-functioning autism) in DSM-IV (see Table 3.2, p. 88-89) currently many people perceive these conditions as indistinguishable, with the children concerned simply representing the high end of a continuum of autistic spectrum disorders (Mayes et al., 2001; Smith, 2004). It is outwith the bounds of this thesis to discuss the minutiae of the debate about whether, and how, HFA differs from AS but one focus of the argument is whether the acquisition of language was delayed or not. Another is whether the presence or absence of movement difficulties might be crucial (see Frith, 1991, for discussion of original papers). In the clinic which referred children for this study, the term HFA was preferred over AS. This might have meant these children were different from those of Green et al. (2002), in terms of early language development. However, since the children were diagnosed on the same tests and seemed comparable on every other measure, it will be interesting to see how they compare on the motor measures included in this study.

A final concern regarding the choice of children to be included in a study of this kind centres on the age range, gender and the choice of a control group. As discussed previously, children's movement competence develops at different rates, along different trajectories and is influenced by many variables. The age group chosen for the present study was 7-10 years so that all participants would have had at least two years in mainstream education but would not yet have reached puberty. Puberty has a major influence on all aspects of development including body dimensions, muscle strength and flexibility. Pubertal changes occur in girls a couple of years earlier than boys therefore at age 9 or10 years some girls will already have significant increases in circulating hormones. For this reason, and in order to try to standardise the sample on as many aspects as possible, other than diagnosis, it was elected to recruit only boys for this study. It is essential to include a control group of children who are developing typically without any motor or other problems. In some studies, two such groups are included, one age matched, the other younger but equivalent on other measures (e.g., Hill et al., 1998). In this study, only a group of boys matched on age and IQ was included.

In order to improve upon previous studies, which have included children from clinical settings, a novel strategy for recruitment and diagnosis was employed. In the DCD field, researchers are usually aware of the participant's diagnosis while assessing the child. This information in itself might bias the experimenter to perceive abnormalities of motor control when there are none. Similarly, if the child is reported to have an attentional difficulty, which is known to the researcher, his/her way of interacting with the child and judgements of performance may be influenced by such prior knowledge. In the present study, effort was made to avoid these problems by single blinding in that the researcher did not know either the source of referral nor diagnosis while administering the assessments. To achieve this, physiotherapy colleagues volunteered to handle all referrals, administer information, exclusion criteria and consent procedures. By reducing researcher bias, this method should increase the reliability and validity of results.

In summary, the final cohort of children assessed 'blind' by the present author comprised three clinical groups, namely, one thought to have DCD, one thought to have HFA (which for comparison purposes are labelled HFA/AS), one thought to have BJHS, plus an age-matched control group of typically developing (TD) children. Although the reduction in the scope of the study was disappointing the comparison between DCD and HFA/AS could be viewed fortuitously, as a replication of earlier work (Green et al., 2002). They compared the severity and nature of motor impairments in two groups of children aged 6-10 years, one with AS and the other with specific developmental disorder of motor function, the WHO label for DCD. In the present study, the addition of a BJHS group was novel and added new information on the movement characteristics of all of the children in the study.

The second design issue to be considered in this investigation concerned the assessments used. In most previous studies, little attempt has been made by researchers to verify that all DSM criteria have actually been fulfilled. For the present study, therefore, it was considered important to build into the assessment process, tests which would help to verify whether each participant met criteria for his diagnostic group i.e., a test with norms that would also ensure that the typically developing group were free from problems. This included a short verbal IQ test, which was available to physiotherapists.

In many studies of children with 'specific' learning difficulties, the assessments are narrowly focussed on one (perhaps primary) domain of function and no attention is given to other factors which are not only important in the process of differential diagnosis but may also confound results. For example, a child may present with DCD but unless other difficulties, which might affect motor competence, are identified, possible interacting effects may be overlooked. An important feature of the present study, therefore, was the inclusion of a range of tests, which would help identify co-existing difficulties. So, while the primary aim was to request and try to obtain participants with a specific unitary diagnosis, which was deemed 'pure' DCD or 'pure' HFA/AS or 'pure' BJHS, an important objective was to measure formally, and document, any co-occurring problems that each child might have.

In order to achieve the objectives just outlined, it was essential to employ the best possible set of assessment instruments at every stage. As a way of operationalising Criterion A for DCD, Study 3 showed that the M-ABC and VMI 3rd edition (Beery, 1989) combined, yielded a success rate of over 70%. Since that study was conducted, the VMI 4th edition (Beery, 1997) was published which includes separate motor and visual perception sub-tests designed to tease out the causes of difficulty with the design copying which comprises the main test component. Instead of using the VMI in the labelling process alone, therefore, in this study, it was also used as a means of identifying children whose motor difficulties might (at least in part) be caused by pure visual perceptual problems. As before, a handwriting test was included in the battery, with some improvements to procedure. For the children referred to the study as High functioning autism or Asperger's Syndrome the high-functioning Autism Spectrum (HFA/AS Screening Questionnaire (ASSQ); Ehlers et al., 1999) was used. This is a brief checklist designed as a supplement to the longer assessment instruments used in this field such as the Autism Diagnostic Interview Revised (ADI-R; Lord, Rutter, & LeCouteur, 1994) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1989). For children referred from rheumatology, the Beighton test (Beighton et al., 1973), a widely used rating of joint hypermobility was used. For all groups, the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997), a short checklist which includes a measure of hyperactivity-inattention in addition to scores on other behavioural, emotional and social domains, was administered. Parents have unique knowledge related to the development of their own child. It was invaluable to tap into their insight and perceptions so they were

asked to provide a background history and to indicate any problem areas including handwriting, reading, speech and language, behaviour and attention.

Having considered the question of whether DCD exists as a separate syndrome, the study then moves to the second question of interest i.e., whether the movement difficulties of children with DCD are the same or different from those of children with other conditions featuring 'clumsiness' as a symptom. In view of the limitations of cluster analysis mentioned in Study 3, the use of this technique was rejected and a different approach was used. In addition to visually inspecting the group profiles of performance on the standardised movement tests employed, the methodology of experimental or neuropsychology was employed to test specific hypotheses about the way children from different groups might respond when motor tasks are varied in particular ways. Although there are numerous studies, which employ this approach to compare children with DCD and TD children, there are few which set out to test specific hypotheses relating to differences between children with movement difficulties from several different diagnostic categories. Consequently, this part of the study was very exploratory.

Briefly, three separate, and rather disparate, lines of enquiry were pursued. The first focussed on possible differences between the groups in response to tasks which require increasingly fine perceptual judgements. Many children with DCD are known to have increasing difficulty as the spatial demands of movement tasks are increased (Volman & Geuze, 1998). In contrast, although children with AS are frequently clumsy, they are often exceptionally good at manual tasks, which are perceptually complex such as completing jigsaws and building models. Similarly, children with BJHS may be challenged to control mechanical degrees of freedom but no one suggests that they have spatial problems. It may well be, therefore, that this area of performance might reveal differences between children with DCD, HFA/AS and BJHS in a way that a global test like the M-ABC would not. For this study, therefore, a specially constructed pegboard was produced which required the children to differentiate between large and small pegs, before inserting them in the board. Specifically, the hypothesis tested proposed that (i) TD boys would have less difficulty than all of the clinical groups with this task, and (ii) that boys with HFA/AS and BJHS would have less difficulty than children with DCD.

The second line of enquiry focussed on possible differences between the groups in their dependence on vision during a simple static balance task. All of us find it harder to balance on one leg with our eyes shut than with them open. However, we hypothesised that hypermobile children might be even more dependent on vision than children with DCD or HFA/AS because they did not receive the same sort of feedback from the joints involved in maintaining balance.

The third line of enquiry pursued was originally designed to separate children with ADHD from the other groups. Recall that these tasks were designed before the study began and the author did not know for some time that children with ADHD were not being referred to the study. The hypothesis here was that children with ADHD would find it much more difficult than the other groups to cope with a secondary task. In this case, a standard dual task procedure was employed and the children were required to count, while completing a pegboard task and a static balance task. In the absence of an ADHD group in the study, the only prediction that we made was that the BJHS and TD group would be less affected by counting than the other two groups who would not differ from each other.

In summary, the aims of this final study were (i) to examine, in further detail, the issues around differentiation of children with DCD from children with other developmental disorders, (ii) to examine any similarities and differences in the motor profiles shown by children with DCD, HFA/AS and BJHS, and (iii) to explore the possibility that subtle differences in motor performance between the three clinical groups could be revealed by specific experimental manipulations.

9.1 Methods and Participants

9.1.1 Summary of Participants, Ethical Approval and informed consent

A total of 51 boys aged between 7 years and 11 years participated in this study. Recruited prospectively from the Greater London area, 39 boys made up the three clinical groups and 12 the control group.

The project was registered with GOSH R & D and was approved by the Research Ethics Committee as a "Multicentre study with no Local Investigator". Local Ethics Committees in each locality area approved the study. The responsible adult for each

child signed the informed consent form (Appendix 6). As the children were all under 11 years, assent forms were made available but were not required.

The procedure and assessment instruments used will be described in two stages: Stage 1 will cover the methodology employed to select subjects; Stage 2 will describe the various tests and measures taken.

9.1.2 Stage 1: Selection of Participants

The aim of this stage of the study was to identify four groups of boys each meeting separate inclusion/exclusion criteria – three clinical groups bearing the labels shown in Table 9.1 and a control group of typically developing children. For comparability, participants in all four groups had to meet the following general criteria:

Male, older than 7 years and younger than 11 years, currently enrolled in mainstream school.

Believed to be of average or above average intelligence.

Understands English.

No current illness or medical problem.

Normal hearing, and vision (with spectacles if necessary).

In addition, boys in all three clinical groups should **not** have received a regular (significant) physiotherapy or occupational therapy movement programme within the last year. In practice the children might come from an out-patient waiting list.

Table 9.1 *Group, Diagnosis, and Main Criteria used*

Group	Diagnosis (or label given).	Main Criteria Used
DCD	Developmental Coordination Disorder DCD	DSM-IV
HFA	High Functioning Autism/Asperger Syndrome/ (HFA/AS)	DSM-IV
BJHS	Benign Joint Hypermobility Syndrome (BJHS)	Beighton score and clinical signs (No DSM-IV guide)
TD	Typical Development	Absence of medical, educational or other concern

The specific criteria applied and procedure employed to obtain each of the three experimental groups, and for the comparison group, was as follows:

Developmental Coordination Disorder (DCD) Group

Boys with possible DCD were recruited from occupational and physiotherapy departments at London hospitals and through community contacts. Referral centres were asked to suggest children whom they considered would meet DSM-IV criteria for DCD e.g., child would have failed a standardised movement test or obtained a score placing them definitely below average on a screening checklist such as the Developmental Coordination Disorder Questionnaire (DCDQ; Wilson et al., 1998), plus clinical judgement. Parent/carers and or teachers should have expressed concern and no medical condition other than 'DCD' should account for the movement difficulty.

High functioning Autism/Asperger (henceforth HFA) Group

As mentioned above boys labelled as HFA were assumed to be the same as those with a diagnosis of AS. Most of the HFA group were recruited from the Behavioural Sciences Unit, Institute of Child Health (ICH), and diagnosed, according to their strict algorithm of outcome on both the 3Di (the Developmental, Dimensional and Diagnostic Interview) (Skuse et al., 2004) and Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1989). The other children came from clinics where a diagnosis of HFA/AS had been given but details of the method of diagnosis was not always specified.

Benign Joint Hypermobility Syndrome (BJHS Group)

Boys with a current diagnosis of Benign Joint Hypermobility Syndrome (BJHS) were recruited from the Hypermobility Clinic organised by the Rheumatology Department at GOSH, and from other medical and therapy clinic sources. At GOSH, a diagnosis of BJHS was made when the child met current clinical criteria (i.e., a Beighton score =/> 6), and exhibited hypermobility during clinical observation. Additionally, it was stressed that the child should not have muscle disease, nor a neurological condition and there should be no active inflammation that might compromise movement assessment. The children should not have been treated for their hypermobility.

Typically Developing (comparison group)

Five mainstream state schools in the London area, were approached for the recruitment of typically developing boys who met the general criteria listed above.

Procedure for recruitment

Recruiting packs were sent/given to identified referral sources. These included a letter requesting referral to the project, an information leaflet, project protocol, consent/assent forms, inclusion/exclusion criteria and additional detail of how to interpret criteria specifically for the project. Fliers were provided where appropriate and worded for a specific group (e.g., hypermobility clinic) (copies in Appendices 3-6).

Procedure for admission to the study

Three physiotherapy colleagues volunteered to receive, record and administer requests for information, make initial contact with families, send out fact sheets and forms, and handle any queries and subsequent referrals in order that the researcher (JMP) who administered the individual assessments would remain blind to the group allocation of each boy. Physiotherapy colleagues allotted each child entered into the project a unique alphanumeric identification code to meet data protection anonymity. Referrals were received en bloc from some sources so to further ensure blinding ID numbering sequence was obtained from a random number web-site (http://www.random.org).

As a result of this exercise a total of 69 children were offered as candidates for inclusion. Further details of the acceptance/rejection of candidates can be found in the results section.

9.1.3 Stage 2: Testing at GOSH

Once contact had been made by colleagues and the family had agreed to take part, the researcher (JMP) was provided with the child's name, ID code, date of birth and contact telephone number in order for the assessment appointment to be arranged. Colleagues sent out appointment confirmation letter with practical details (suitable clothing including trainers, directions to GOSH etc). Additionally, unless there was a definite medical contra-indication, families were requested not to give their child any medication on the day of appointment and in the case of Methylphenidate (Ritalin) or similar drugs to stop for at least three days beforehand. At the appointed time the

family arrived at GOSH physiotherapy department for the assessment and was met by the researcher (JMP). The assessment lasted between 2-3 hours and was completed on a single morning or afternoon visit.

The present researcher administered all tests except the British Picture Vocabulary Scale (BPVS-II). All assessments took place in the same quiet room in the physiotherapy department at GOSH. Parents and the child were reminded once again at the start that the researcher did not know the source of referral and that they should refrain from giving such information. They were reassured that at the end of the session there would be opportunity and time to ask questions and talk freely. Parents chose whether to remain in the room for the entire assessment (up to three hours) or to leave during part of the time. A certificate was awarded to the child at the end of the assessment and travel expenses were immediately refunded.

The assessment was divided into two sessions with a break for refreshment (juice and biscuits) at a suitable mid-point. Since the tests and measures employed in these two parts of the study were quite different in their format, they too will be described separately.

Part 1. Standardised tests and clinical observation

The first part of the assessment comprised a series of tests, some completed by the child, others by the parents. They ranged from fully standardised instruments to much less formal checklists and clinical observations. After a brief chat to establish a relaxed interaction with the child, six cards with pictures to illustrate the main assessment tasks were laid out in front of the child. In order to allow the child to take some control, he was asked to choose which task he would like to start with. His preferred choice was recorded. Following this first task, the order of items was not standardised but followed what appeared to be the best sequence to maintain his interest and attention. Parents completed their components either in the room or in the adjacent waiting area. Details of each test and the procedure adopted are given below.

Tests completed by the child

British Picture Vocabulary Scales (BPVS-II; Dunn et al., 1997)

Purpose: The aim of the BPVS-II is to assess a child's receptive English vocabulary and as such is often taken as an index of general intelligence. It is a widely used test for children aged, 3 to 15 years. It requires no reading, speaking or writing, only a simple pointing response to pictures.

Procedure: Administration takes under 10 minutes. The tester says a stimulus word and the child is instructed to point to the appropriate picture from four plates on each page. The child is given practice on two training plates to ensure that the instructions and procedure are understood. The child starts at the set of plates corresponding to his age and works through progressively more difficult vocabulary until his ceiling is reached.

Scoring and psychometric detail: The number of correct responses are recorded, and converted to standard (Mean = 100 *SD* = 15) or percentile scores. The test was standardised in 1995 on 2571 UK children. The manual provides extensive reliability, validity and bibliographic data.

Study 5 Comment: The test was presented according to the manual instructions, by one of the pre-trained physiotherapy volunteer coordinators.

Movement Assessment Battery for Children (M-ABC; Henderson & Sugden, 1992)

Study 5 comment: Details of the M-ABC were provided in Study 3. The test was administered as described on pps 33-89 in the manual. As participants were aged 7-10 years age-bands 2 (7/8 years) or 3 (9/10) were used.

Developmental Test of Visual Motor Integration–Revised Fourth Edition (VMI; Beery, 1997)

Purpose: The stated aim of this test is to identify, children and students aged 3-18 years who may need special assistance. The most recent version of the test comprises three components, one identical to the original and two supplementary components, one designed to measure the visual perception skills required to perform the main test, the other designed to assess control of the hand independent of the perceptual element.

Procedure: A developmental sequence of 27 geometric forms is copied with paper and pencil. In the supplemental visual perception test the child chooses one geometric form that is exactly the same as each stimulus from among others that are not identical. As many as possible of the 27 stimuli are to be identified in three minutes. For the motor coordination section the child traces the stimulus forms with a pencil without going outside the double-lined paths.

Scoring and psychometric detail: One point is awarded for each correct item with a maximum score of 27 on each section. Raw scores are converted to standard scores (Mean = 100, *SD* = 15) and percentiles are provided. The VMI was standardised on 2614 children in USA in 1995-6. Detailed psychometric information is provided in the manual.

Study 5 comment: The VMI 4th edition was administered according to the manual. All children completed the visual perception sub-test, which followed the main test component. In view of the length of the assessment battery and the inclusion of other similar manual dexterity tests (e.g., Item 3 of the M-ABC) the motor supplementary test was administered only for children who demonstrated obvious failure on the main VMI test.

Grapho-motor Tests

The handwriting assessment was similar to that used in Study 3. The items were presented in a standard order as follows:

Task 1 Sentence copying, best: the child was asked to copy 'the quick brown fox jumps over the lazy dog' in their 'best' handwriting. The child was unobtrusively timed as the first letter started and stopped when the last letter was completed.

Task 2 Sentence copying, fast. The child was asked to copy the same sentence as many times as possible in two minutes. This was openly timed using a stopwatch.

Scoring: Handwriting proficiency was measured in two ways. In the absence of an objective method of scoring the quality of handwriting, a simple rating procedure was adopted for the purpose of this study. First, since handwriting is a taught skill which changes as children progress through school, the 51 handwriting samples were divided into two sets, one containing boys age 7/8 and one containing boys 9/10

years. The two age-sets were then randomised and presented to two experienced primary school teachers starting with the young age bundle and immediately followed by the 9/10 year set. They were asked to put the samples into two piles, one which they rated as 'good' hand writing relevant to the child's age and the other which was considered as 'poor'. It was emphasised that the numbers of samples in each group did not have to be equal. Raters were specifically asked to attend to the 'motor' aspect/execution of writing rather than mark a child down because a word was mis-spelt or omitted. The aim was to gather an overall impression of which samples suggested evidence of a movement problem. Agreement between the two raters was moderate (kappa .35). As in Study 3, a quantitative measure of speed of production was also calculated but in the present study, there is space only to present the qualitative judgements made by the teachers.

Anthropomorphical measures

The child's height, weight (kg) and arm span (cms) were measured when standing bare foot, wearing shorts/tracksuit and T-shirt. Body Mass Index (BMI) was calculated (Weight kg/height m^2) and the results compared with published charts (Cole et al., 2000). These were drawn from an international survey from Brazil, Great Britain, Hong Kong, the Netherlands, Singapore and USA and comprised a total sample of 192,727 subjects (male and female) aged from birth to age 25 years. The authors include BMI scores indicating 'overweight' or 'obesity' for boys aged 2-18 in 6 monthly intervals.

Hypermobility or excess muscle and joint flexibility was then measured using the method described by Beighton (1973). The Beighton test is a nine-point screening test requiring that five manoeuvres be assessed, four passive movements which the assessor carries out and one action performed by the subject:

1. Passively dorsiflex the fifth metacarpophalangeal joint to $\geq$ 90 degrees. (R & L)
2. Passively oppose the thumb to the flexor surface of the forearm (R & L)
3. Passively hyperextend the elbow to =/> 10 degrees. (R & L)
4. Passively hyperextend the knee to =/> 10 degrees (R & L)
5. Actively bend forwards to place hands flat on the floor with arms straight.

Right (R) and left (L) scores for the passive items are summed and added to the active item to give a maximum total score of 9. The criteria for children are presently undergoing further reliability studies and should be interpreted with caution

(Grahame et al., 2000, p. 1778). Recent validation of the Beighton test in Dutch children aged 4-12 years recommended a cut off point of ≥ 5 (van der Giessen et al, 2001). Maillard and Murray (2003, p. 35) comment in regard to children that "There is no universally accepted Beighton score.... Some have taken 5/9 others 6/9." However a report of joint laxity in a cohort of 1845 Swedish school children aged 9-15 years recommended a higher cut point which takes both age and gender into consideration (Jansson, Saartok, Werner & Renström, 2004). The study found that a cut off of ≥ 4 used in some studies would result in every second girl and 40% of boys in the Swedish sample as falling into the hypermobile range! As a result of their findings, Jansson et al. (2004) suggest that at age 9 boys and girls should only be considered as hypermobile with a Beighton score of ≥ 8 (the nearest score identifying the highest 5% of their study population for that age group). Supplemental clinical observations such as excess hip range, winging of scapulae, presence of bruising, ability to touch nose with tongue tip (Gorlin's sign), history of dislocated joints or fractured bones are also often included in clinical practice to support the identification of hypermobility. In addition to the problems of scoring, the Beighton test has been criticised for inclusion of only a few joints and that it does not reflect the precise degree of hypermobility, but in its favour it is both quick to administer and provides a general clinical guide (Bulbena et al., 1992).

Parent-completed Questionnaires

Following an explanation of their format, the four questionnaires detailed below were completed by parents (Appendices 8-11). The researcher did not see the completed questionnaires until after the assessment was finished. The two published and two novel questionnaires were piloted on 10 children attending clinic for assessment. Pilot parents completed the questionnaires without difficulty and the scores reflected the clinical observation made by the therapist.

Autism Spectrum Screening Questionnaire (ASSQ; Ehlers et al., 1999).

Purpose: The high-functioning ASSQ is a 27 item checklist suitable for completion by non-professionals for identification of high-functioning autism spectrum disorders/Asperger syndrome in children and adolescents with normal intelligence.

Procedure: The ASSQ takes less than 10 minutes to complete. Twenty-seven statements on whether 'this child stands out as different from other children of

his/her age' are rated on a 3-point Likert scale (0 = No; 1 = Somewhat; 2 = Yes). Examples include 'is regarded as an 'eccentric professor'; 'lives somewhat in a world of his/her own with restricted idiosyncratic intellectual interests', 'wishes to be sociable but fails to make relationships with peers'; 'has special routines: insists on no change'.

Scoring and psychometric detail: Raw scores are summed to give a total score (range 0-54). Cut points for the ASSQ when rated by parents are: $\geq$ 19 'autistic spectrum'; 13-18 'socially impaired'; $\leq$ 12 'normal range'. Good reliability and validity data for this questionnaire are published (Ehlers et al., 1999; Campbell, 2005). Recent research on a total population of 9430 children aged 7-9 years supported the concept of autism as a spectrum. The authors estimate that a total of 5.6% of children in the total population would score $\geq$ 15 on either the teacher or parent ASSQ and recommend a lower cut off be used for screening (Posserud et al., 2006).

Study 5 comment: The parent-completed cut point of $\geq$ 19 was adopted but the recent suggestion by Posserud et al. (2006) that this may be too strict was taken into account for very borderline scores.

Strength and Difficulties Questionnaire (SDQ; Goodman, 1997)

Purpose: The SDQ is a brief measure of the prosocial behaviour and psychopathology of 3-16 years olds that can be completed by parents, teachers or youths.

Procedure: The 25-item questionnaire is quick to administer. It uses a similar Likert-like format to the ASSQ outlined above. Examples of items include: 'Restless, overactive, cannot stay still for long', 'Many fears, easily scared', 'Often lies or cheats', 'Rather solitary, tends to play alone'.

Scoring and psychometric details: The SDQ generates scores in five domains of psychological adjustment: prosocial behaviour; peer problems, hyperactivity-inattention; conduct; emotional difficulties. All but the prosocial sub-scores are summed to generate the Total. The 'normal', 'borderline' or 'abnormal' ranges of scores for the SDQ (parent completed) are as follows:

	Normal	Borderline	Abnormal
Total Range 0-40	0-13	14-16	17-40
Prosocial	6-10	5	0-4
Peer Problem	0-2	3	4-10
Hyperactivity	0-5	6	7-10
Conduct	0-2	3	4-10
Emotional	0-3	4	5-10

The items selected for the domains were based primarily on DSM-IV diagnoses such as ADD, ADHD, AS and Conduct Disorder (CD) (APA, 1994). Normative data from a British sample of 10,438 children aged between 5 and 15 years is accessible on the web (www.sdqinfo). Good reliability and validity have been reported (Goodman & Scott, 1999; Goodman, 2001; Goodman et al., 2000). The SDQ has been used as a screening tool for child psychiatric disorders in UK (Goodman et al., 2000).

Study 5 comment: In the present study the parent/teacher version for children ages 4-10 years was completed.

Parent-rated coordination/flexibility questionnaire

Aim: Ten statements, in a similar format to those of the SDQ but specifically related to coordination (5) and flexibility (5), were designed by the present researcher for the current study.

Procedure: Parents were asked to mark their response as either 'Not true', 'Somewhat true' or 'Certainly true'. The statements included 'Looks awkward when running, hopping or skipping', 'Has broken or fractured a bone' 'Muscles and joints are very flexible, mobile (seem 'double jointed')' 'Often complains of aches and pains in limbs'.

Scoring. Scores were 0, 1, 2 for each item as for the SDQ. Total *s*core range was from 0 = no problem to a maximum of 10 for flexibility items and 10 for coordination items.

Main Problem Area Questionnaire

Parents rated a list of eight common problem domains. The domains included the following: 'Speech & language'; 'Concentration', 'Overactivity', 'Movement & Coordination', 'Relationships & Friendships'; 'Reading', 'Obsessive Interests &

Routines', 'Angry Outbursts'. They were asked to rate/order the items from 1–8 with 1 indicating the greatest problem area and 8 the least. A score of 0 was awarded for any areas which they did not considered to be problematic.

Other tests/data not reported here

Several measures were employed in this study, which will not be reported here. An objective measure of muscle strength was obtained for each child using a hand-held dynamometer to assess grip with the preferred and non preferred hand. Each child was also asked to draw a picture of a person on plain paper. During these tests the researcher recorded the child's hand grip by circling pre-prepared drawings of hand postures. Additional qualitative observations on performance, behaviour and relevant comments made by the child were also recorded.

Part 2. Experimental tests and measures

The second part of the assessment included manual dexterity and balance tests devised by the researcher. Each element included a dual task which involved the child counting while either moving pegs or balancing. Prior to presenting these tasks, therefore, an estimate of the child's counting ability was made informally as a 'game'. The tester, in a chatty manner, usually while clearing away manipulables said to the child "Can you show me how you count in 3s?" If this was deemed too challenging the examiner said "How about 2s?" The aim was to find the level that demanded the child carry out the task cognitively rather than produce an over learnt automatic skill.

The three manual dexterity tasks, using pegboards were presented first, in a standard order. Items 1 and 2 (using the original M-ABC pegboard) were followed by Item 3 using the modified pegboard. At the completion of the pegboard tasks the child took a rest if he wished following which the three balance items were presented in a standard order 1-3. Rest or free activity (stretching, jumping) up to one minute between each task was allowed if the child wished.

Pegboard item 1 (turning pegs): The M-ABC pegboard with 16 small pegs in the holes was placed on the M-ABC mat on the table directly in front of the child. The child was asked to invert each small bi-coloured peg using only one hand. After a practice trial including just four pegs, each hand was tested and timed.

Pegboard item 2 (turning pegs + counting)

Item one procedure was repeated but this time the child completed the task while counting in 3s, (2s or back wards in 1s if there was difficulty). Counting was recorded on a tape recorder. The tape recorder was started at the word 'go' and simultaneously with starting the stopwatch. The stopwatch was stopped as the child released the last peg.

Pegboard item 3 (inserting variable size pegs)

The specially designed pegboard with alternate small and large holes was placed directly in front of the child. A small box containing eight large and eight small pegs, was placed on the same side of the board as the child's hand that was being used for the task. The aim of the task was to place all pegs into the appropriate size holes, one at a time in any order, as fast as possible. A practice attempt consisting of one row of pegs was provided. (The tester unobtrusively marked the sequence of peg-placement on a pre-prepared sheet of paper during the task but these data are not reported here).

Balance item 1 (Baseline standing balance R and L)

The child was asked to balance on each leg for as long as possible up to 30 seconds. He was allowed to start with either leg. Arm movement was permitted but hooking one leg around or against the other, was not. The time that the child was able to balance without shifting the supporting foot from its start position was recorded.

Balance item 2 (Standing balance R and L + counting)

The child repeated balance task 1 while counting. The tape recorder was started simultaneously with the "go" command and stopped if the child shifted his foot from the start position or when he reached 30 seconds. The child chose which leg to start on. Both legs were recorded.

Balance item 3 (Standing balance R and L eyes closed)

Child repeated balance item 1 with eyes closed.

Equipment

Pegboard and pegs. The M-ABC pegboard and small pegs were used as a template for a technician to modify the board design and pegs to include different sized holes

and pegs. Eight holes and matching pegs retained the M-ABC measurements. Alternate holes on the board (eight) were re-designed measuring 5mm diameter and eight pegs constructed from wooden doweling to fit these holes. All pegs were 20mm in height. One side of the pegboard was identified by placing a strip of yellow tape on its edge in order to facilitate a standard orientation of large/small holes. A shallow cardboard box lid 80x55x15mm was used to contain the pegs.

Sony Cassette Recorder

A Sony Cassette Recorder (Model TCM-200D/150 Sony Corporation) with external unidirectional boundary microphone (Model YOGA HM-0026) were used to verify that the child was counting during the manual dexterity and balance items in Part 2 of the assessment procedure. The voice operated recording mode was switched off during recording. This ensured that recording continued during hesitations or silent periods.

9.1.4 Closure of Assessment

At the completion of the assessment, before scoring any tests formally, the researcher used her clinical observation of the child to decide and record to which referral group (DCD / HFA / BJHS / TD) the child belonged. This was then discussed with parents and they divulged to the researcher the source of referral and the child's diagnosis if any. Following a short discussion of any difficulties that they felt the child had, the four questionnaires completed by the parent were collected in.

Background developmental history questionnaire, including face sheet data and details of any information relating to intervention and Special Educational Needs Code of Practice (DES, 1981; DFE, 1988; 1994; DfES, 2001) was completed by the parent and sent to the volunteer colleagues as part of the recruitment procedure (Appendix 7). This questionnaire together with any additional information given to colleagues were subsequently delivered to the researcher. Following calculation of all scores, a written assessment summary and brief comments was sent to the parent and referrer.

9.1.5 Data collected and analysis

A summary of the data recorded from each of the tests is shown in Tables 9.3-9.7.

Data were analysed using SPSS for Windows version 12.0 (Norussis, 2005). A combination of parametric and non-parametric tests were used.

9.2 Results and Discussion

9.2.1 Finding children is not so easy!

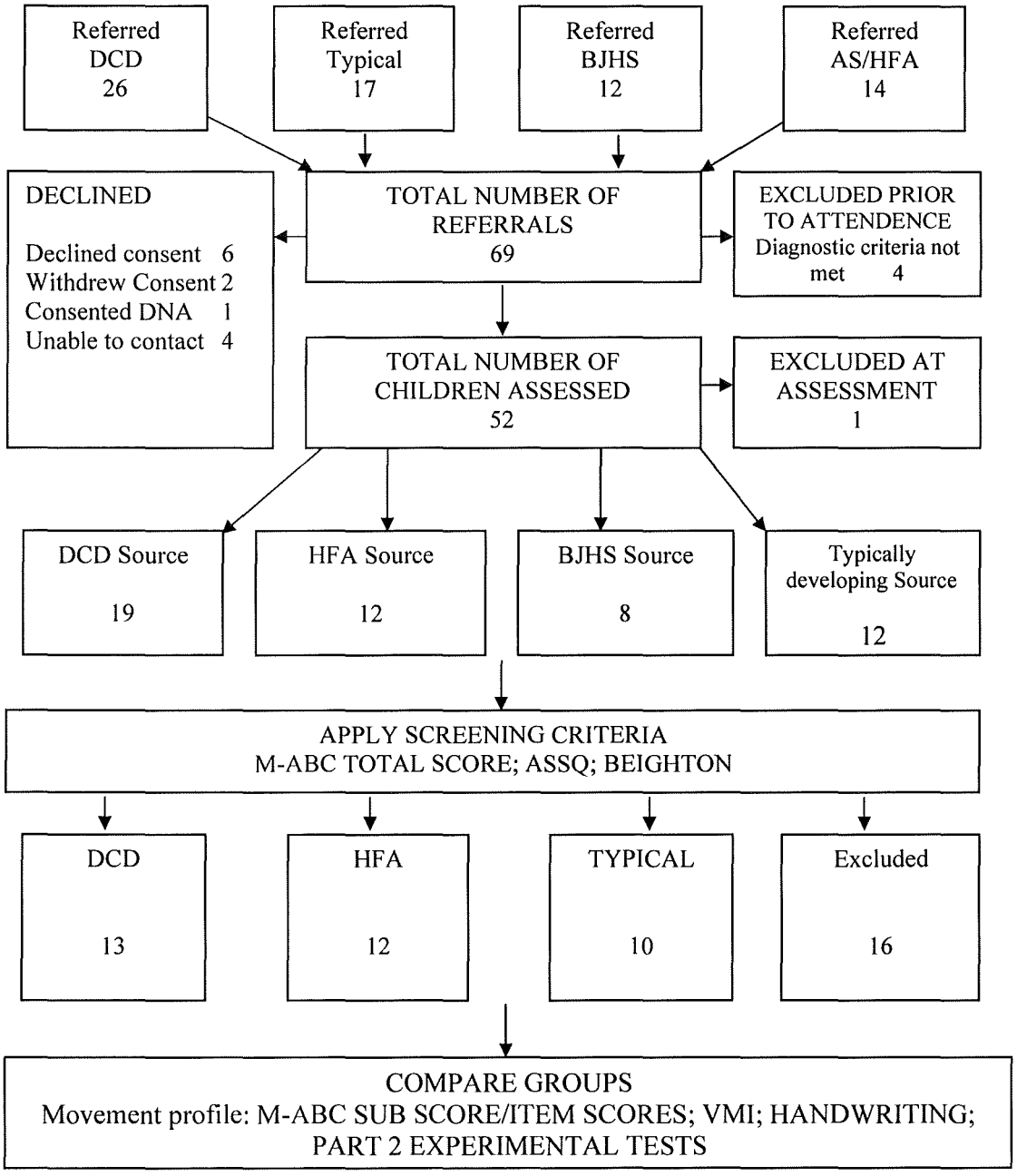
The original objective in this study had been to find six groups of 15 children, five clinical groups and one control group. In the end, however, only four groups were identified and in three of these the numbers were fewer than planned. Although suitable specialist sources for the recruitment of children with ADHD and Tourette's (TS) were contacted and ethical approval was agreed, no children were referred. Interestingly, one of the main reasons given was that the referrers were unable to identify boys with only one of the conditions requested. For instance, referrers commented that at least two thirds of children with TS also meet criteria for AD/HD. Similarly, very many children with ADHD demonstrate tics or fully diagnosed TS. In addition, there was the problem that there is a dearth of volunteers for research and available participants are at a premium and precious! The TS clinic was currently running several research projects and children at GOSH are restricted in the number of studies in which they are allowed to participate. This is monitored through a computerised Patient Information Monitoring System (PIMS). From the five mainstream primary schools approached three declined with one spending several weeks with the protocol details then sent the message that 'as the school had been busy supporting the Tsunami disaster they felt they could not take on anything more!'

There were other possible reasons for the failure to reach the targets set for recruitment in this study. One was the time-scale. Obtaining ethical approval within the NHS is a rigorous process often taking several months in a single centre. In a multicentre trial such as this one, each centre had its own protocol, which had to be followed, thus delaying progress even more. Another problem might have been the GOSH consent form (Appendix 6). This is a standard form which outlines certain rights and indemnities but the 'small print' is daunting and may raise concerns that make a parent hesitant to sign. While the 'blinding' process was viewed as a very positive feature of this study, it may have compromised recruitment by removing any direct contact between the potential participants and the person they would meet at assessment. Finally, London was on high security alert for terrorist activity during the period of the study.

Figure 9.1 shows a flow chart of the recruitment of subjects, attrition, assignment to groups etc. Sixty-nine boys were referred to the project: 26 DCD, 14 HFA, 12 BJHS and 17 TD. Thirteen of these families either could not be contacted or declined to take part at various points in time (see Figure 9.1). One boy was excluded from the study because he was too old and three were excluded on medical grounds. The latter included a child diagnosed with G6PD deficiency (an enzyme deficiency disorder predisposing to haemolytic anaemia), premature birth and kernicterus and hearing loss, a child with Cornelia de Langue and Raymonds syndrome and complex seizures medicated with Epilim and a child with a history of periventricular leucomalacia, epilepsy, ADHD and high functioning autism. Finally, one child attended the assessment but it soon became apparent that he should be excluded. He had scored on the 2nd percentile on the BPVS-II and his extensive difficulties were supported both by the clinical impression of the researcher and parent report.

In summary, the initial protocol for the study estimated a cohort of up to 75 boys and although 69 were referred the final number of children who consented to take part and met the stringent inclusion criteria was 51.

Figure 9.1 *Flow Chart of Recruitment, Attrition and Assignment to Groups*



9.2.2 Characteristics of the participants and comparability of the groups on non-motor factors

The primary objective in this study was to compare the perceptuo-motor performance of the four groups of participating children. Ideally, therefore, the aim was to find groups of boys who were fairly comparable on non-motor aspects such as age, cognitive ability and body build. A total of 51 boys assented to take part and all completed the full assessment between January 2004 and June 2005. Table 9.2 shows means and SDs for age, verbal IQ, height, weight, BMI and arm span for the four

groups of boys before diagnosis had been formally checked. One-way ANOVAS on these data revealed no significant differences between groups. Results of the BMI showed that seven children (14%) fell into the 'overweight' category. However, no child rated as 'obese' (Cole et al., 2000) and there was nothing to suggest that the overweight children were confined to only one group.

Table 9.2 *Non-motor Characteristics of Groups*

	DCD	HFA	BJHS	TD	F	Stats. Sig (p)
Age (months)	103 (16)	104 (14)	107 (16)	105 (13)	.166	.919
BPVS (IQ)	109 (13.7)	100 (16.7)	112 (15.1)	109 (13.6)	1.136	.345
Height (cm)	132 (7.0)	133 (9.1)	133 (11.0)	135 (8.9)	.380	.768
Weight (kg)	28.0 (5.7)	28.1 (5.8)	31.1 (8.7)	30.8 (8.5)	.692	.561
Body Mass Index (BMI)	16.2 (2.4)	15.8 (1.9)	17.2 (2.5)	16.6 (3.0)	.657	.583
Arm span (cm)	131 (8.0)	133 (9.7)	133 (12.3)	136 (12.4)	.499	.685

Code: Mean (SD) and F value
DCD: Developmental Coordination Disorder, HFA: High Functioning Autism, BJHS: Benign Joint Hypermobility Syndrome, TD Typically Developing

9.2.3 How satisfactory was the control group?

The purpose of a control group (TD) is to act as a reference against which experimental groups may be compared. The control group, for this study, comprised 12 children referred from mainstream London schools, who were thought to demonstrate typical development and therefore, would be expected to fall within the normal range on all of the standardised measures included. Table 9.3 shows the data for this group on all measures outlined above. As the table shows, 10 boys had completed the BPVS and all scored above 80. The two others were judged by their teachers to be of average IQ. On the Movement ABC, no child fell below the 5th percentile but there were two boys who scored below the 15 $^{th.}$ (see below for further discussion of these two boys). On the Beighton Scale, one boy with a low M-ABC score had a borderline score and complained of muscle aches. Two others were definitely hypermobile but had no associated problems. None of these boys came close to meeting criteria for problems on the ASSQ or SDQ.

Table 9.3 *Typically Developing Referral Group*

Case	Movement Measures				Measures in other domains			Parent Information
	Total M-ABC (%ile)	Total VMI (%ile)	Handwriting Problem		ASSQ Total	Beighton	SDQ Hyper-activity	Labels listed by parents plus details of any other problems including whether reading (R) or speech and language (SL) noted as a difficulty by parents/teachers.
			Teacher	Parent				
40TD	96	73	No	No	0	4	4	No problems
47TD	89	61	No	No	0	4	2	No problems
42TD	49	45	No	No	3	5	4	No problems
43TD	18	84	No	No	4	5	0	No problems
41TD	79	25	No	No	0	6	3	No problems
49TD	79	65	No	No	0	2	2	Colour blind red/green No other problems
50TD	70	68	No	Yes	2	1	3	R problem and had help with handwriting. SL OK.
48TD	89	23	Yes	Yes	No data	5	No data	Extra help for literacy and reading
51TD	13 #	50	No	Yes	3	6	3	Coordination problem. No R or SL problems..
45TD	13 #	34	No	No	6	7 #	0	Complains of muscle aches. No R or SL problems.
44TD	96	55	No	No	1	9 *	3	No problems
46TD	96	37	No	No	2	9 *	1	No problems

*Abnormal range # Borderline

Handwriting: teacher - handwriting rated good or poor. Parent - handwriting rated as a problem = yes ; no problem = no.

Although probably unnecessary, a statistical check on the differences between the control group and the three clinical groups was carried out. The latter were combined and t-tests were performed on all relevant variables. As Table 9.4 indicates, the differences between the two groups were highly significant on all but the Beighton flexibility score, an issue which will be discussed in more detail below.

Table 9.4 *Experimental Groups and Control Group t-Test Results*

Test	Experimental Group $n = 39$ Mean (Range)	Control Group $n = 12$ Mean (Range)	t
M-ABC Total score	14.6 (2-36)	3.6 (0-10.5)	4.47**
Full VMI (Standard Score)	90.3 (72-120)	100.75 (89-115)	-3.25**
ASSQ[a] Total score	16.1 (0-46)	1.9 (0-6)	3.69**
SDQ[a] Total score	15.9 (1-7)	4.4 (1-30)	5.35**
Beighton Total score	5.7 (0-9)	5.2 (1-9)	.550 ns.

[a] One parent failed to complete forms. Stats: ** $p < .01$

Legend: M-ABC: Movement Assessment Battery for Children, VMI: Developmental Test of Visual Motor Integration, ASSQ: Autism Spectrum Screening Questionnaire, SDQ: Strengths and Difficulties Questionnaire, Beighton: nine-point hypermobility score.

9.2.4 Who fits in what box?

As noted above, one of the objectives of this study was to obtain three distinct groups of children, each from a different source. Great care was taken to describe to the referrers the kind of child required for the study. The aim then was to examine the children's scores on a series of objective measures chosen specifically to support or reject the referral diagnosis and to explore any overlap between the emergent groups. Although the 'inclusion' assessment instruments used for each group might not have been as comprehensive as one would have liked, all were tried and tested and had been used in other research studies. As far as exclusionary assessments were concerned, this study faced the same problem as any other. It is simply not possible to test for every problem that children with 'specific learning difficulties' might exhibit. While the SDQ does cover a fairly wide range of difficulties that children

commonly experience, for others the necessary information was extracted from checklists and comments documented by parents. Although much more data is available and will be reported elsewhere, in what follows, the focus of attention will be on the boys' scores on the M-ABC, VMI, and selected aspects of the handwriting test in the motor domain, and on the Beighton, ASSQ, and selected aspects of the SDQ and parent checklists in other domains.

As might be predicted from preceding studies in this thesis, the fit between the diagnosis expected from the referral source and the actual picture presented by each child was not perfect. Since the issues that arose for each group of children were different, each referral group will be dealt with separately before presenting the picture as a whole. For each group, the order of presentation of test scores reflects the diagnostic category being discussed.

DCD Group

Eighteen of the 19 referrals for the DCD group came from an OT or PT clinical waiting list for assessment and one child was referred directly from school via a special educational needs coordinator (SENCO). Although screening on a movement test had been suggested it became evident that this had rarely taken place and referral to the study was generally based on information provided by parents or medical professionals which the experienced OT or PT interpreted as suitable. This process contrasted markedly with procedures adopted by the clinic referring children with HFA/AS for this study (see below).

Table 9.5 summarises the data obtained for these 19 boys on both motor and non-motor measures. As noted earlier, discussion regarding diagnostic criteria for DCD is inconclusive. However, scores on the M-ABC are frequently used as at least one of the criteria for identification. Which cut-off point is chosen, however, leads to criticism either that the 5th percentile may be overly strict or the 15th percentile too lenient. If one takes the M-ABC score as the **primary** means of applying Criterion A for DCD the results in this study, indicate that out of the 19 boys referred as 'DCD' 13 met this criterion with eight obtaining scores at or below the 5th percentile (indicated by a * and dark shading) and five scoring below the 15th (indicated by # and light shading). If, however, the VMI is taken as an alternative/addition to the M-ABC, the results show that a further 3 children would be included, taking the total to

16 (interestingly, two of these children scored below the 25th percentile on the M-ABC and only one showed a marked discrepancy between his scores on the two tests). Examination of the remaining three boy's data showed that two scored below the 25th percentile on the M-ABC and had difficulty with handwriting as judged by their parents. For one child, however, a problem with handwriting was reported by the parent but not picked up in the teacher rating. In summary, by shifting the operationalisation of Criterion A considerably from being broad-based but strict (M-ABC 5th percentile) to being highly specific and lenient (a global rating of handwriting), in the end, it allowed account to be made for the referral of every child in the group. Moreover, when the parent's comments were examined in detail, most, but not all, were aware of the movement difficulties their child experienced.

Table 9.5 *DCD Referral group*

Case	Motor measures				Measures other domains			Parent Information
	M-ABC %ile	Total VMI %ile	Handwriting problem?		ASSQ Total Score	Beigh ton Total Score	SDQ Hyper-activity	Labels listed by parents plus details of any other problems including whether reading (R) or speech and language (SL) noted as a difficulty by parents/teachers..
			Teacher	Parent				
13 DC	1 *	55	No	Yes	2	3	5	Moderately dyslexic. Cousin autistic. R & Writing problem. SL?
15 DC	1 *	21	Yes	Yes	9	2	6 #	School raised handwriting coordination problem. SL NAP.
7 DC	1 *	19	No	Yes	7	6	5	Reading & Coord. problem. SL NAP. Sister dyslexic
4 DC	1 *	30	Yes	Yes	7	6	3	Reading & SL NAP. Coord. problem.
14 DC	1 *	9 #	Yes	Yes	13 #	7#	8 *	Convulsion (MMR). Reading NAP. SL & Coord prob. Brothers dyslexic
17 DC	5 *	23	Yes	Yes	5	5	6 #	Readng & Coord? problem. Relative dyslexic and dyspraxic.
3 DC	5 *	30	No	Yes	6	2	7 *	Teacher and parent report slow reading. SL and Coord NAP.
6 DC	5 *	19	Yes	Yes	7	1	10 *	ADHD ? dyspraxia. R & Coord. problem. SL ? prob. Dad ?dyslexic.
12 DC	6 #	70	Yes	Yes	16 #	4	9 *	Reading & SL NAP. Coord. roblem. Cousin dyspraxic
2 DC	7 #	7 #	No	Yes	20 *	9*	8 *	Reading & SL problems.
16 DC	8 #	25	Yes	Yes	10	8*	7 *	Mother and brother dyslexic. Reading, SL. NAP.
8 DC	11 #	13 #	Yes	Yes	0	2	6 #	Was v. flexible. Mother flexible. R. & SL NAP. Coord problem
9 DC	15 #	27	Yes	Yes	0	8*	0	Reading, SL and Coordination NAP.
5 DC	70	4 *	Yes	Yes	7	6	7 *	Reading problem. SL NAP.
18 DC	16	9 #	No	Yes	23 *	8*	5	Mother thought child a bit 'autistic'. Coord problem. SL NAP.
11 DC	20	14 #	No	Yes	23 *	9*	2	Dyslexic. Mother? dyslex. R. & coord prob. SL NAP. 'Growing pains'.
19 DC	16	91	Yes	Yes	28 *	8*	6 #	Reading problem. SL NAP.
10 DC	26	50	No	Yes	25 *	8*	9 *	"Loose joints". AS not confirmed.. SL & Coord. Problem. R. NAP
1 DC	22	55	Yes	Yes	6	6	5	Reading and SL NAP. Coordination problem.

* Abnormal score # Borderline score NAP = Not a problem.

The next question to be addressed was how many of these children had isolated movement difficulties (i.e., might be described as 'pure' cases of DCD) and how many had difficulties in other domains. In some cases, associated difficulties might mean that the child met criteria for another condition. Other difficulties might simply be problems that arose as a result of being 'clumsy'. Even the briefest of inspections of Table 9.5 would suggest that many of these children had other problems, which concerned their parents. Examination of the ASSQ scores for the 19 DCD referrals, revealed that five boys were clearly above the cut-off score of 19 for HFA and a further two fell in the borderline "socially impaired" range. Thus, seven children (37%) referred from the 'DCD' source might well have had HFA/AS or at least be on the autistic spectrum, which on current DSM-IV criteria excludes a diagnosis of DCD. In two of these cases, the family suspected HFA or the child had been assessed briefly but a diagnosis on the autistic spectrum was never confirmed.

As noted above, the Beighton measure for hypermobility has not been fully validated for children and there is considerable debate about the most appropriate cut-off point for definite identification. In view of the greater degree and prevalence of flexibility in young children (Jansson et al., 2004) a stringent criteria of $\geq$ than 8 was used as the 'definite' cut-off for hypermobility and 7 as 'borderline' in this study. Of the 19 children in this group, eight were in the definite or borderline hypermobility range. Four of these children had M-ABC scores below the 15th percentile and four did not.

If one extends enquiry beyond the four main tests used in this study to include components of the SDQ score, such as the element relating to hyperactivity and/or parent report of a reading or language difficulty, six children score in the abnormal range leaving only three 'pure' DCD cases.

HFA Group

Twelve children were referred to the study believed to have HFA. Eight children were referred from the same highly regarded specialist clinic following identification of HFA using the same algorithm derived from assessment on the 3Di plus the ADOS. Two of the other four children were diagnosed at the same hospital but by different consultants using similar but not identical assessment and two were given their diagnosis elsewhere.

Table 9.6 shows results for this group, with dark and light shading indicating abnormal and borderline scores. On the ASSQ, nine boys scored above 19, thus clearly confirming the accuracy of their initial diagnosis. Two boys scored between 17 and 19 and in view of recent debate about the scoring of the ASSQ (Posserud et al., 2006) they too were considered to meet inclusion criteria for HFA for this study. The remaining boy scored 12 (just below the ASSQ borderline) and although he met 3Di and ADOS criteria it was felt that his motor difficulties might be influencing function to the detriment of social scores rather than the other way round.

Table 9.6 *High Functioning Autism (HFA) Referral Group*

Case	AS/HFA Measure	Measures in Other Domains						Parent Information
	ASSQ Total Score	Total M-ABC (%ile)	Total VMI (%ile)	Handwriting problem? Teacher	Handwriting problem? Parent	Beighton Total	SDQ Hyper-activity	Labels listed by parents plus details of any other problems including whether reading (R) or speech and language (SL) noted as a difficulty by parents/teachers.
29AS	46 *	1 *	23	No	Yes	6	7 *	Autism. Reading problem.
37AS	37 *	16	50	No	Yes	8 *	9 *	Social problems. Reading not a problem.
30AS	36 *	1 *	30	Yes	No	3	6 #	Semantic pragmatic disorder/AS.Infantile spasms. Reading NAP. SL problem.
38AS	32 *	1 *	19	Yes	Yes	5	7 *	HFA. Dyspraxic. Reading problem.
28AS	28 *	1 *	3 *	Yes	Yes	6	7 *	Atypical seizures (not now). HFA. Reading NAP. SL problem
34AS	24 *	79	18	Yes	Yes	6	10 *	ADHD on Ritalin. Reading problem. SL NAP.
36AS	24 *	9 #	23	Yes	No	6	7 *	AS. Reading NAP.
33AS	23 *	32	63	No	Yes	8 *	7 *	AS. Reading and SL problem.
35AS	21 *	1 *	30	Yes	Yes	7 #	8 *	AS. Reading problem. SL NAP.
32AS	18 #	1 *	5 *	Yes	Yes	6	10 *	Hydrochephalus as a baby. HFA. Reading problem.
39AS	17 #	6 #	47	Yes	Yes	4	4	Mild HFA. Brother 'double jointed'. Reading NAP. SL and coordination problem.
31AS	12	2 *	50	No	Yes	2	8 *	AS. Reading, SL, NAP. Coordination problem

* Abnormal score # borderline score NAP = Not a problem

To determine which, if any, of these 12 boys had a substantial movement difficulty, scores on the M-ABC and VMI were then examined. Nine boys failed the M-ABC: seven scored below the 5th percentile and two below the 15th. A tenth boy scored on the 16th percentile and had difficulty with handwriting. The remaining two boys had M-ABC scores on the 32 and 79th percentile but both were reported to have handwriting difficulties by their parents. In contrast to their scores on the M-ABC, only two children in this group failed the VMI full test (both below the 5th percentile) and both of these had low M-ABC scores. As in the DCD group, handwriting difficulties were common, with the parents reporting difficulties in all but two boys and teachers concurring in most cases. The question of how these results compare to those reported recently by Green et al. (2002) will be dealt with later. However, it is worth noting here that although **every** child in their small sample failed the M-ABC, these results are not dissimilar.

Three children in this group were hypermobile (two definite and one borderline). As with the DCD group, however, hypermobility scores did not seem to be closely related to scores on the M-ABC in that the borderline child had a very low M-ABC score, and the two definite children passed.

Hyperactivity was marked in this referral group with 11 boys scoring in the abnormal range on the SDQ hyperactivity sub-section, one borderline and only one boy (who just failed the ASSQ) scoring in the normal range. One child who gained a ceiling hyperactivity score had actually been given a label of ADHD (as well as HFA) and was medicated with Ritalin although this was stopped several days before his assessment for the present study.

In summary from the HFA referral group, 11 of the 12 boys clearly met criteria for HFA and one was questionable. Interestingly, this boy scored very poorly on the M-ABC (2nd percentile) and his parents commented at the closure of the assessment that they had always been rather more concerned by their son's movement difficulty than his lack of social skills which had always been the focus of intervention. As with the DCD referral group, hyperactivity was commonly reported by parents in this group as were difficulties with reading, language etc. Had a more comprehensive/ objective test for ADHD been included, then some of the HFA as well as the DCD might have qualified for this diagnosis too.

BJHS Group

The referral route for the BJHS was from a hypermobility clinic attached to a Rheumatology clinic in GOSH. As noted above, BJHS is normally defined as hypermobility accompanied by aches and pains in joints and/or muscles. As symptoms are highly subjective and may vary from week to week, however, a clear algorithm for diagnosis does not exist. Indeed, the situation with regard to differential diagnosis is even less clear than it is for DCD. Since the children referred to this study had been tested on the Beighton Scale already as part of a clinical examination by highly trained clinicians, however, it was assumed that the referrals were accurate.

Table 9.7 presents data on the eight BJHS referrals. When these boys were tested on the Beighton Scale using the standardised procedures we had developed, four children were given scores of 8, two had scores of 7 and three scored 6. Surprisingly, although referred from the hypermobility clinic specifically for the project, one boy demonstrated markedly **stiff** joints with a Beighton score of 0 (confirmed by a specialist orthopaedic physiotherapist as second rater). The child's parents commented that he had attended physiotherapy for stretching of tight muscles at one clinic and physiotherapy for BJHS at another centre! Since there is so much uncertainty about the interpretation of scores on this test (one point lies between abnormality and borderline), and no reliability data that we know of, this raises several questions regarding the fit between our assessment and that of the referring clinic. It is possible that the experienced clinicians assessing these children were picking up symptoms, such as flexibility in other parts of the body which are not addressed in the narrow focus of the Beighton Scale. This might mean that we should apply a lenient criterion in this study and include all children scoring above 6, say. However, if we appeal to the small amount of literature on this topic then we would only consider those children scoring 7 or more as meeting criterion for BJHS. In summary, with a lenient criterion, we would give the label BJHS to seven of the eight children, with a stricter criterion, only four out of eight would be eligible.

Table 9.7 *Benign Joint Hypermobility (BJHS Referral) Group*

Case	Flexibility measure	Measures in Other Domains						Parent Information
	Beighton Score	Total M-ABC (%ile)	Total VMI (%ile)	Handwriting problem?		ASSQ Total Score	SDQ hyper-activity subtest	Labels listed by parents plus details of any other problems including whether reading (R) or speech and language (SL) noted as a difficulty by parents/teachers.
				Teacher	Parent			
26BJ	8 *	36	16	Yes	Yes	4	8.5 *	Dyslexic ADHD Dyspraxic BJHS + relative Tourette's
27BJ	8 *	2 *	45	Yes	Yes	15 #	5	EDS hypermobile type (son and mother). Coordination problem. R and SL problem.
24BJ	7 #	5 *	10 #	Yes	No	1	7 *	No R or SL problem
21BJ	7 #	1 *	3 *	Yes	Yes	43 *	10 *	Autistic tendency, CP, dyspraxia. Coordination problem
25BJ	6	9 #	25	No	No	0	1	No R or SL problem. Writing fingers go 'numb'
20BJ	6	9 #	27	Yes	Yes	3	9 *	Dyslexia queried. Reading prob.. SL NAP. Dad possibly dyslexic
23BJ	6	20	47	No	No	1	2	No R or SL problem.
22BJ	0	79	27	Yes	Yes	29 *	10 *	BJHS ADHD HFA(?) but not diagnosed. Reading prob.

* Abnormal score # borderline score NAP = Not a problem

Beighton Flexible 8-9 Borderline 7 Pass ≤ 6

Various recent studies have commented on the fact that many children with BJHS are also 'clumsy' (Russek, 1999; Maillard & Murray, 2003; Murray, 2006; Kirby et al., 2005) and this was confirmed by parent report in this study. As Table 9.6 shows, three of the eight boys obtained M-ABC scores below the 5th percentile and two below the 15th percentile. Four of these five boys failed the VMI and/or had handwriting difficulties that were noted by both parent and teacher. There were, however, three boys who passed both the M-ABC and VMI, one of whom was hypermobile and one not at all.

On the ASSQ, two boys scored in the HFA range and one scored as 'socially impaired' (borderline HFA). Two of these children were also below the 15th percentile on the M-ABC. Five children in the group scored in the hyperactive range and for two of these, parents wrote that the label ADHD had been suggested at some stage. Both children who fell well into the HFA range on the ASSQ had had a tentative label related to the autistic spectrum applied to them.

In summary, from the original eight children referred as BJHS, no child actually had 'pure' joint hypermobility. Two boys meet criteria for HFA and one additional child fell within the 'socially impaired' category. Three other boys had definite or borderline movement difficulty commensurate with DCD and one boy although scoring in the normal range on all other tests was below the 25th percentile on the M-ABC. Children with BJHS are commonly described as fidgety and constantly moving and making postural adjustments. As noted in the other groups a large proportion of the group were hyperactive.

9.2.5 Pure cases, co-occurrence and co-morbidity - where do we stand now?

Under the headings "who fits into what box?" and "how good is the control group?", each of the four groups of referred children was described in detail. What these descriptive sections showed was that the referral systems employed for each group varied considerably and had a very definite effect on the type of child being referred. Some children very clearly fitted into the diagnostic category being proposed, whereas others did not. For example, among the children who were referred as children with HFA, the referral system used was very detailed, which meant that the tests used in this study tended to confirm the diagnosis originally made. In contrast,

the BJHS clinic was clearly less precise in the precedures they used, the outcome being that the fit with the tests used in this study was less good. This fit/misfit notwithstanding, however, what the data showed were that by using a wider range of tests than might otherwise be employed, various difficulties that were common to all groups were revealed. To some extent, the validity of these observations was supported by the fact that such problems were almost entirely absent in the control group.

In Chapter 3, a hypothetical diagram (Figure 3.1) showing possible overlap between various developmental conditions is presented. What becomes very apparent from the real data yielded by this study is that the picture is just as complex as others have suggested (see e.g., Kaplan et al., 1998). What is more, these data show that the picture can be made to change considerably in that classification of individuals and therefore group boundaries change according to the criteria employed and the cut-off points selected. Consider the following. Unlike many studies involving children with DCD, this study began with 3 'clinical' groups of children, rather than just a DCD plus control group. All children were tested on standardised tests, which were well established in each of the three areas of concern and which were administered under identical conditions. Because **all** children had been tested on all tests this provided the opportunity to examine overlap in some detail. Even if one employs the children's scores on the standardised tests alone (i.e., the M-ABC and/or VMI, the ASSQ, and the Beighton) one can see how each group changes as a function of whether one uses the 5th or the 15th percentile as a cut off point in each case. These changes are illustrated graphically in Figures 9.2a and 9.2b, with the data shown in Figure 9.2a illustrating the overlap when the 15th percentile or more lenient criteria are adopted and Figure 9.2b when the 5th percentile or stricter criteria are selected.

It can be seen that from the original cohort of 51 boys, applying the stricter criteria (5th percentile or higher cut point for ASSQ and Beighton), 35 met criteria for one or more of the target conditions. Twenty (57%) met criteria for just one condition, fifteen (43%) two conditions but no child met criteria for all three conditions. The corresponding figures when less strict criteria are adopted revealed 41 participants met criteria for one or more target conditions. In 18 (44%) instances this was a single condition; for 16 (39%) criteria were met for two conditions and for seven children (17%) criteria were met for all three conditions. Thus between 43% and 56% of the

children meet criteria for at least two conditions on whichever current criteria are selected. Moreover, it has already been noted above that if we had incorporated into this analysis, some of the other test data available from this study, the picture would have changed yet again.

In summary, the data reported here adds yet more solid, empirical support for the idea that there is considerable overlap between many of the childhood disorders, which are often treated as if they were easily separable. For example, the present results were rather similar to those of Kaplan et al. (1998) who looked at the overlap between DCD, ADHD and reading difficulty (dyslexia). When they used the 15th percentile cut off, 46% of children emerged with a unitary condition, 54% with two conditions and 20% with all three conditions co-occurring. Now, this study has added HFA/AS and BJHS to the set, the latter having been previously thought to be more medical in origin than the others. What is not clear from any study, which collects data at one point in time, however, is whether the picture obtained would have been the same some years before, or indeed, some years after. This was one reason why, for example, data on behaviour problems were not included in this analysis. A child with a movement difficulty might have consistently co-ocurring difficulties in these domains, but equally likely, the behaviour problem might be the result of the way the movement problem was dealt with by the family and school. From a purely practical point of view, however, these data emphasise the need to extend the range of assessments used in all children's settings as widely as possible, while at the same time considering time, cost and issues of expertise.

Figure 9.2a *Overlap of DCD, HFA and BJHS using 15th percentile or borderline cut off (n = 41)*

Typically developing (n = 10)

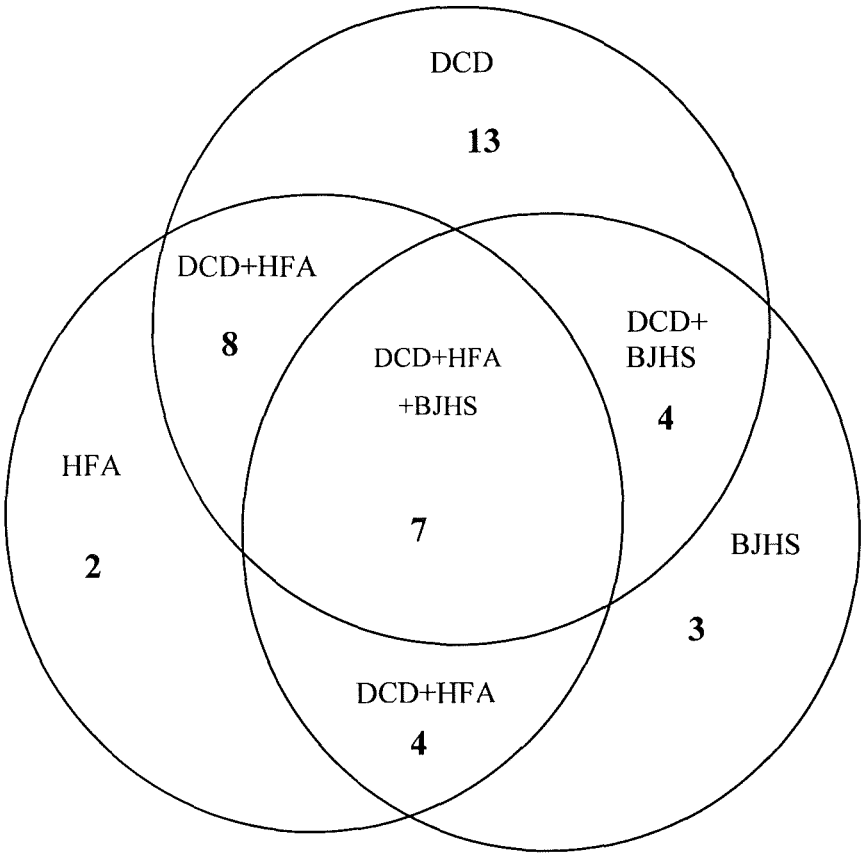
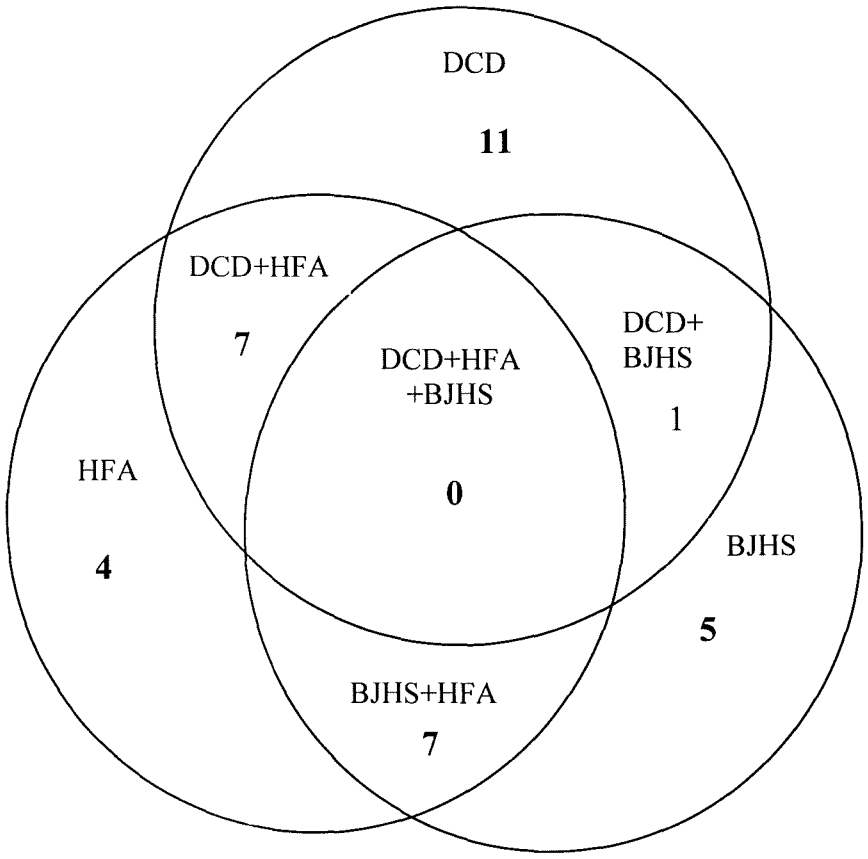


Figure 9.2b *Overlap of DCD, HFA and BJHS using 5th percentile or stricter cut off (n = 35)*

Typically developing (*n* = 16)



9.2.6 Conclusion

This section detailed the recruitment and methodology adopted for the first part of the study. The hurdles faced in attempting to find and recruit 'pure' cases were related to disparity in the concepts and variable algorithms and procedures used by the referrers. The breadth of the assessment battery adopted for the study revealed that many of the children demonstrate overlapping features rather than fitting neatly into a diagnostic box. The overlap is dependent upon many factors not least of which are the cut points chosen for 'caseness'. However the battery was successful in separating out a minority of children who do appear to have well defined 'pure' movement difficulty compatible with the DSM-IV diagnosis of DCD, without co-occurring features of medical pathology or symptoms within the autistic spectrum.

There were others who met criteria for HFA who had movement problems. The next section turns to the question of whether the 'clumsiness' apparent in differing diagnostic groups such as DCD and HFA is peculiar in some way to one or other group or whether the 'clumsy ' feature is just a general cluster of motor symptoms common to each.

9.3 Movement Difficulties in selected clinical groups– same or different?

The primary focus of this thesis is DCD and the movement problem that is its core feature. Although the overlap between diagnostic categories as a whole is an important issue, the final question now addressed in this study is whether the **movement** difficulties experienced by children from different diagnostic groups, differ and in what way. To address this question, we begin by reorganising the 51 referred children into groups according to their movement competence, then proceed to examine the profiles of their performance on selected motor and perceptual measures. Last, but not least, we test the specific hypotheses set out in Section 9.0 employing the experimental tasks outlined on page 228-9.

Originally, it had been hoped to find children with DCD, children with HFA/AS who were 'clumsy' and children with BJHS who were also 'clumsy' (as well as children with ADHD who were clumsy). . Having looked at the tests originally believed to yield objective and reliable data, however, the difficulty with the Beighton Test, now shown to be difficult to score and unreliable in children (Jansson et al., 2004; Ferrari et al., 2005; Murray, 2006) arose once again. To take account of this problem, it was therefore decided to reorganise the boys for this part of the study in two ways. First, participants would be grouped on the more reliable measures, the M-ABC, VMI and ASSQ scores using a lenient cut-off at the 15th percentile and ignoring the Beighton results. Second, a regrouping using the Beighton test but cutting across the original groups would be undertaken but treated with more caution than the previous grouping.

Re-examining the data shown in Tables 9.3-9.7 for the original groups, using the M-ABC, VMI and ASSQ produced the following groups: All but two children from the original TD group remained in the final group (TDf). Two boys with M-ABC scores just below the 15th percentile were excluded because neither failed the VMI or had

been perceived as having motor problems by an adult. Thus, they were deemed not to meet Criterion B for DCD.

From the original DCD referral group, 10 of the 19 boys failed the M-ABC and passed the ASSQ and therefore met criteria for inclusion in the final DCD group (DCDf); Three moved into the final HFA group (HFA/ASf) - 1 failed the ASSQ and the M-ABC; 2 failed the ASSQ and VMI, were below the 25th percentile on the M-ABC and had poor handwriting. The remaining six children were excluded because they passed the movement tests or had complex comorbidity.

From the original HFA group, eight boys failed the ASSQ and the M-ABC and thus met criteria for HFA with movement difficulty (HFAf). Four were excluded: three passed the M-ABC and one scored in the 'socially impaired category' with movement difficulty (to have included the last boy in the DCD final group would 'muddy' the attempt to keep all groups as 'pure' as possible).

From the original BJHS group, three failed the M-ABC but passed the ASSQ therefore met criteria for the DCD final group (DCDf). One boy failed both the M-ABC & ASSQ and was moved into the HFAf group. Four children were excluded: two passed the M-ABC, VMI and ASSQ, one failed the M-ABC but was in the 'socially impaired' range, and one failed the ASSQ but passed the M-ABC.

On the second pass through the data, two additional sub-groups of children with movement problems were identified: a 'motor-difficulty-hypermobile' (MD-hyp) sub-group of boys who failed the Beighton (score $\geq$ 7) **and** the M-ABC ($\leq$ 15th percentile) and a group who failed the M-ABC but were **not** hypermobile (MD-nonhyp). These groups comprised 11 and 20 boys respectively. Finally, of interest in the context of the Green et al. study (2002) who reported all their AS children to fail their motor tests, boys who met Criteria for HFA yet passed the M-ABC were identified as an 'HFA-no motor problem' sub-group.

In summary, from the original 51 referrals, ten children met criteria for inclusion in the typically developing comparison group (TDf), 13 children met criteria for a 'pure' diagnosis of DCD (DCDf), and 12 met criteria for HFA with a motor difficulty

(HFAf). When hypermobility was used as a defining feature, 11 boys with and 20 boys without flexibility were identified (MD-hyp/nonhyp).

9.3.1 Comparability of the final groups of boys

Table 9.8 shows means and SDs for the DCDf, HFAf and TDf groups for age at assessment, body mass index (BMI), height, weight, and IQ. ANOVA on age and anthropomorphic measures revealed no significant differences between the three groups (max $F = 1.7$). In contrast, ANOVA on the BPVS-II scores indicated that the main effect of group was significant, $F(2,28) = 10.5$, $p < .001$). Pair-wise comparisons between the three groups then showed that the boys in the HFAf group had significantly lower scores than either the DCDf or TDf boys ($p < 01$ and $p < .05$ respectively) but the DCDf and TDf did not differ from each other.[4]

Table 9.8 *Group means and SDs for age, anthropomorphic measures and IQ*

	DCDf	HFAf	TDf	MD-hyp	MD-nonhyp
Age (months)	104 (15)	102 (16)	105 (11)	98.8 (17.0)	104 (14.0)
BMI	17.0 (2.5)	15.4 (1.6)	17.1 (3.1)	15.4 (1.5)	16.3 (2.4)
Height (cms)	133 (8)	130 (10)	135 (8)	129 (10)	133 (7.9)
Weight (kg)	29.9 (6.6)	26.2 (6.4)	31.8 (8.8)	26.1 (5.8)	28.7 (6.3)
BPVS-II Standard Score	112 (8)	94.4 (10.5)	107 (11)	101 (11.2)	110 (16.8)

MD-hyp = Motor-difficulty-hypermobile

MD-nonhyp = Motor-difficulty-non-hypermobile

Identical analyses for boys with and without hypermobility showed no significant difference on any variable (see Table 9.9).

Profiles of performance on the standardised motor tests

Table 9.9 shows the means and SDs for the five new groups of boys on the Movement ABC total scores along with its three components, the VMI main test and the visual sub-test component, plus an estimate of parent's perception of whether a child had handwriting difficulty.

[4] A few of the following analyses were repeated employing BPVS score as a covariate but this had no effect on the outcome.

Table 9.9 *Group means and SDs for M-ABC, VMI and Handwriting rating*

	DCDf (13)	HFAf (12)	TDf (10)	MD-hyp (11)	MD-nonhyp (20)
M-ABC total score	15.1 (4.9)	20.5 (9.6)	2.2 (2.8)	14.5 (5.9)	18.4 (7.7)
M-ABC manual dexterity sub-score	7.0 (3.5)	8.5 (5.1)	1.0 (1.4)	6.5 (4.0)	7.9 (4.1)
M-ABC ball skills sub-score	3.3 (2.7)	5.9 (3.1)	0.2 (0)	3.5 (3.6)	5.1 (2.8)
M-ABC balance sub-score	4.8 (3.3)	6.1 (4.5)	1.0 (1.9)	4.6 (3.5)	5.4 (3.8)
VMI main test Standard score	89.5 (5.0)	84.1 (8.7)	102 (8)	85.5 (8)	90.6 (8.5)
VMI visual sub-test Standard score	104 (11)	97.8 (21.5)	111 (14)	93.9 (17.7)	108 (15.1)
Handwriting difficulty Parent perception	10 (77%)	10 (83%)	2 (20%)	9 (82%)	15 (75%)

Beginning with the M-ABC and its component parts, ANOVAs on these data revealed significant differences between the DCDf, HFAf and TDf groups on the total scores as well as on all sections (Total Score $F(2,32) = 22.3$; Manual Dexterity $F(2,32) = 12.3$; Ball skills $F(2,32) = 14.8$; Balance $F(2,32) = 6.2$, (min $p = <.01$). As the table shows, these differences were largely accounted for by the difference between the TD group and the two clinical groups. Of particular interest at this stage, however, were the differences, if any, between the DCDf and HFAf groups. Although there was a general tendency for the HFAf group to score less well on each component, post hoc tests on each of these sub-scores showed that only the ball skills reached statistical significance ($p < .05$) a finding consistent with that of Green et al. (2002). Interestingly, too, if one looked at the six HFA boys who were excluded because they did **not** fail the M-ABC/VMI screen, four were unable to catch competently although none failed the age appropriate aiming task.

On the M-ABC and its component parts, identical analyses showed that there were no differences on any measure between the MD-hyp and MD-nonhyp groups (minimum $p = .3$).

On the VMI main test, ANOVA on the scores obtained by the DCDf, HFAf and TDf groups revealed a main effect of group, $F(2,32) = 15.4$, $p < .01$) but once again this

effect was due to the superiority of the TD group. A post hoc test of the difference was between the HFA and DCD groups was not significant (p = .21, *ns*) Similarly, the TD group stood out from both the MD-hyp and MD-nonhyp but these two groups did not differ from each other.

On the visual perception component of the VMI, the main effect of group was not significant for the comparison between DCDf, HFAf and TDf, $F(2,32) = 1.8$, *ns*) but there was a main effect of group for the comparison between the TDf, MD-hyp and MD-nonhyp groups, $F(2,38) = 3.9$, $p < .05$). On this component, the DCD children had slightly **higher** scores than the HFA but this difference was not significant. Conversely, there was a tendency for children who were hypermobile to have **lower** scores than those who were not hypermobile (p = .06).

Of particular interest as far as the clinical groups were concerned was whether failure on the two components of the VMI were linked. Examination of the individual subject data revealed that of the two children from the DCD group, who failed the main test, neither failed the visual element. Within the HFA group, the comparable figures were six failures on the main test with only two failing the visual component. Three children failed only the visual component, but two of these were actually below the 25th percentile on the main test and the third was on the 30th. While these data suggest that the addition of the supplementary visual and motor components to the original VMI helps to untangle subtle differences in this complex perceptual-motor domain, there are actually very few children whose difficulty with copying can be accounted for by the fact that they do not recognise the shapes to be copied. Most children who have difficulty on the main test may have a problem in motor execution, but more likely the difficulty lies in integrating visual and motor processes (see Wedell, 1972). That said, there are a few children whose failure on the main test seems likely to be caused by a specific visual-perceptual problem, a pattern not uncommon in children born prematurely (Jongmans et al., 1998). In the present study just one boy was born prematurely (< 28 weeks gestation). His scores on the VMI main and visual component were 3rd and 30th percentile respectively. In addition, it should be remembered that the VMI visual test is a timed test with the children forced to complete the matching of designs within 3 minutes. The visual component is always performed after the main test and some children with attention difficulty may lose concentration.

A similar analysis of the MD-hyp and MD-nonhyp groups showed that 8 (73%) of the MD-hyp children failed one (4) or both (2) tests. In contrast, 5 (25%) of the MD-nonhyp group failed the main (2) or the visual (2) test but none failed both components. These results suggest that hypermobility may be an area for further research in relation to visual perception and motor function.

Parents' perceptions of handwriting as a problem for their child showed no difference between the two clinical groups with each judging around 80% to have a problem as compared to 20% of the TDf children ($\chi^2(2) = 11.3, p < .01$.).

In summary, although the DCD and HFA groups in this study were very different from each other in many ways, as far as their movement profiles on the standardised tests were concerned, there was very little difference between them - except perhaps on ball skill. This pattern of results is very similar to that obtained by Green et al. (2002) who also used the M-ABC in their study (see below for further discussion). There were no differences between the children who failed the M-ABC and were judged to be hypermobile on the Beighton test and those who also failed the M-ABC but were not hypermobile. The lack of any significant result may reflect the true picture but may again reflect the lack of reliability of the Beighton. Further examination with much larger groups and a standardised measure of connective tissue morphology would be a valuable future study.

9.3.2 Experimental tests: Alter the task, alter the response?

After exploring the group profiles on standardised measures, a quite different approach to searching for differences between the clinical groups was taken. As a general principle, this involved altering the demands of selected movement tasks in such a way that possible differential effects on specific groups of children could be explored. In order to take as broad a view as possible at this stage, it was elected to sample one task from the fine motor domain and one from the gross motor domain. For consistency with the M-ABC content, the fine motor task selected was peg manipulation and the gross motor, static balance. Three ways of changing task demands were chosen - increasing perceptual complexity, removing vision and adding a secondary task. Within each of these mini experiments, specific hypotheses were tested. As noted above, however, there was little experimental data to go on in

most cases. While the number of studies involving DCD and TD children has grown, studies comparing one clinical group with another are few.

The experimental tests were completed after the break for refreshment and in most instances the children were keen to carry on and enjoyed the new activities. However, there were a few who were less cooperative, or tired and failed to complete one or more of the seven tasks. For example, one child from the HFAf group had required so much encouragement from his mother and the researcher to complete the first half of the assessment, it seemed pointless to continue with the experimental items; another three children from this group did not complete all items; and one child from the TDf group did not attempt the balance items at all. From the DCDf group there was just one score missing due to technical reasons.

Results of the experimental tests

In the following section the observations, results and statistical analyses are presented with each mini-experiment or task, being presented separately. Group means and SDs are given in the tables and/or figures accompanying each section and individual data are included in Appendix 13. Although the children completed all three tasks with both their preferred and non-preferred arm and leg, data are presented for the preferred only. In addition to the fact that there was no evidence of differential effects due to hand/leg used, the problems associated with interpreting effects due to crossed or mixed laterality suggested another thesis might be required to deal with the data! (see Hiscock & Chapieski, 2004, for a comprehensive review).

The effect of increasing perceptual complexity on speed of hand function

It is often said that children with AS or HFA are exceptionally good at making models or completing puzzles and there are numerous case studies to support this view. Such comments are rarely made about children with DCD. In fact, quite the opposite, parents often report, that their child hated any game that involved making perceptual judgements and avoided things like LEGO when little.

There is considerable support in the literature for the idea that some, but not all children with DCD have a visual- perceptual problem which might play a part in their difficulty "making the hand do what the eye sees" (Hulme et al., 1984; Henderson et al., 1994). We could find no studies, which suggested this was the case

in children with HFA children. For this component of the study, therefore, we hypothesised that the DCD children would have more difficulty with the uneven-sized peg task than the HFA children, and consequently would be much slower. However against this prediction, we did consider the possibility that the children with HFA might focus on the detail of the pegs themselves, the opportunity for pattern making and give less priority to speed. With less assurance, we also predicted that the hypermobile children would find it hard to feel the difference between the peg diameter and resort to checking the pegs visually. In turn, this would slow them down more than the non-hypermobile children.

Qualitative Observation

On this task, most children immediately noticed the difference between the fat and thin pegs when the novel board was presented with its uneven sized holes. When moving uneven pegs some children picked out all one size peg first, others chose a peg to fit the next sequenced hole and others picked a peg at random and found a hole to match it. Children also varied in their strategy when placing pegs. Sixteen (50%) sequenced the pegs in lines, four either completed the central section of the board then the perimeter or placed pegs in diagonals. These last four children all came from the clinical groups (DCDf 2; HFAf 2) and all were in the MD-nonhyp sub group. Both DCDf children had completed the M-ABC peg task in the usual manner but both HFAf boys had an odd approach also to their M-ABC peg task: one was haphazard in moving rows and hopped over pegs blocking his path and the other interrupted peg placing after every four pegs to walk to the window and look out. Seven (70%) of the TDf groups used a sequence strategy. One child recited 'fat' 'skinny' as he placed each different peg which appeared to be an organisational strategy. However, there was nothing to suggest that any of these strategies were peculiar to one or other of the clinical groups.

Quantitative data.

Table 9.10 shows the means and SDs for all the five groups of children. ANOVA on these data approached significance for the main effect of group, $F(2,30) = 2.9$, $p = .07$). Post hoc tests then showed that the TDf-HFAf comparison was the only comparison to even approach significance ($p = .07$). Contrary to our prediction, there was a slight tendency for the HFA children to be slower than the DCD but this was

not significant. Similarly, the difference between the MD-hyp and MD-nonhyp went in the opposite direction to our prediction but was not significant.

Table 9.10 *Means and SDs for all five groups on uneven peg placing task*

Item	DCDf	HFAf	TDf	MD-hyp	MD-nonhyp
Uneven peg place preferred hand (in seconds)	47.1 (12)	55 (14.7)	42.2 (8.9)	45.8 (10.4)	51.0 (14.4)

Why might this be so? First, the fact that some but not all of the DCD children in the present study failed either the VMI main task alone or the VMI plus the visual component supported existing studies of children in this group, suggested that differences **within** the groups might outweigh differences between them. Also, what became evident when we analysed the standardised test results was that some but not all of the HFA children had the same problems, suggesting that generalisations about their 'good' perceptual abilities were not accurate either. In order to explore these data a little further, individual subject data were inspected and the relationships between various test scores examined. This showed, for example, that there was no correlation between uneven peg placing and either the main VMI or VMI visual component (Pearson Correlation r = -.192 and .130 respectively). Once again, therefore, we are faced with the fact that tasks, which appear to share certain characteristics, such as high visual complexity do not correlate with each other. In this group of children, the uneven peg turning correlated significantly with the child's M-ABC Manual Dexterity score (Pearson correlation $r = .48$, $p = .005$) and also with the peg turn task with preferred hand from the experimental set of tests ($r = .608$, $p < .001$), suggesting that finger dexterity and speed of movement might play a more important role in this task than size judgements.

In sum, contrary to prediction, this task revealed no differences between any of the clinical groups involved. The TD children all completed the task faster than the DCD or HFA children and seemed to organise their actions more systematically. Between the DCD and HFA children, nothing was found to separate the two either quantitatively or qualitatively. This was true also of the MD-hyp and MD-nonhyp groups. What stood out instead were the individual differences which crossed all diagnostic categories.

Balance task: Effect of removing vision

Postural control or balance underpins all of the actions we make. This can extend from standing on one leg to put on a pair of trousers to sitting upright when writing. The importance of balance for movement competence is reflected in the numerous research studies that investigate the development of static balance including aging issues from various perspectives (Clark & Watkins, 1984; Shumway-Cook & Woolacott, 1985; Williams & Ho, 2004). Similarly, problems with balance in children (and in the elderly) has been the focus of many other studies (Wann et al., 1998; Geuze, 2003; Riach & Starkes, 1993; Forseth & Sigmundsson, 2003; Alderson et al., 2006). In parallel, the importance of balance as a crucial component of competence is reflected in the many motor proficiency assessment batteries which contain items to evaluate both static and dynamic balance, often labelled postural control and locomotor ability (Barnett & Peters, 2004).

All of us find it harder to balance on one leg - or even on two- when we close our eyes. This universal observation lies at the heart of many studies, which set out to investigate the role of vision in maintaining balance. For instance, Geuze (2003), Forseth and Sigmundsson (2003), and Wann et al. (1998), all examined static balance in children of primary school age with movement problems and included tasks with and without vision. All studies, although their inclusion criteria and aims were different, reported poorer balance when vision was occluded for all participants. Also, children with movement problems had more difficulty with the balance tasks than the normal comparison group, but there were, of course, individual differences within the DCD group. Studies reviewed in Chapter 2 discussed several studies which suggested that a sub-group of DCD children with specific balance difficulty may exist (Wann et al., 1998; Jongmans, 1993). However, these studies do not tell us the precise nature of the problem, and there are no equivalent studies of other clinical groups of children. Consequently, they do not help us with a specific hypothesis to test, regarding a differential effect of removing vision in a static balance task, that might emerge between groups from different diagnostic categories. In the present study, therefore, we predicted that all groups would find it harder to balance with their eyes closed but that the effect would be larger for the DCD and HFA groups who would not differ from each other. A prediction that we felt more confident about, however, concerned the effect of hypermobility on balance. Due to their ligamentous laxity resulting in increased degrees of freedom throughout the body

mechanics possibly sensory feedback would be more variable and demand for reinforcing muscle co-contraction greater. Thus it was predicted that the MD-hyp would be more affected by the removal of vision than children who were not hypermobile.

Observation: balance with and without vision

Balance items were completed barefoot and stabilising adjustments in the muscles around the ankle and small muscles of the foot were clearly visible. Some children swayed the head and trunk or made gross arm movements while attempting to remain balanced. Some attempted to grasp onto furniture, hold onto their foot, hook the leg etc and had to be reminded that none of these was allowed. For the second part of this test children were asked to close their eyes tight shut and as far as possible the researcher tried to ensure that there was no peeking. The addition of a blindfold had been considered but rejected in view of the effect that this could have on the child's behaviour.

Results

The data for the TD children and the two pairs of clinical groups are shown in Figures 9.3a and 9.3b.

Results for the TDf DCDf HFAf groups two way ANOVA revealed that both main effects were significant, the main effect of group, $F(2) = 6.5$, $p = .01$) and of vision/no vision, $F(1) = 76.8$, $p < .001$). However, contrary to prediction the interaction between group and experimental manipulation was not significant ($p = 0.2$). Pair-wise comparisons were significant for the TDf-DCD groups ($p = .01$) and TDf-HFAf groups ($p = .02$) but the DCDf-HFAf was not.

Figure 9.3a *Balance with and without vision for the DCDf, HFAf and TDF groups*

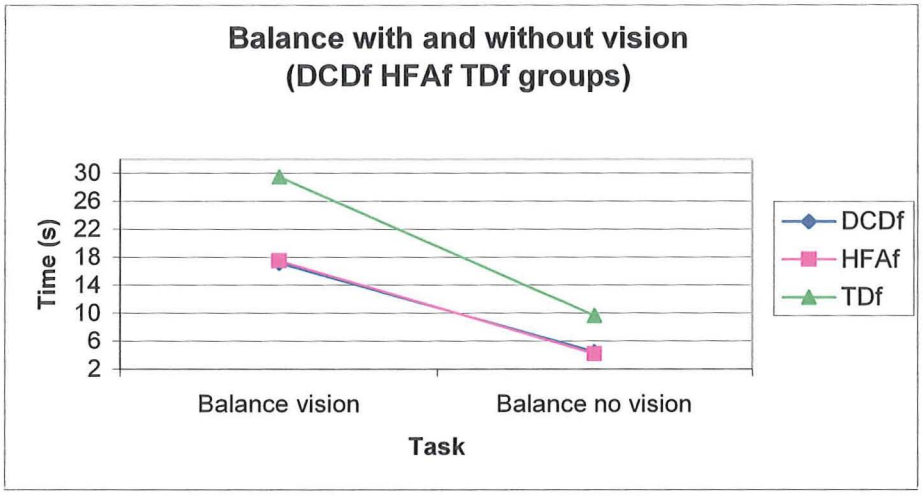
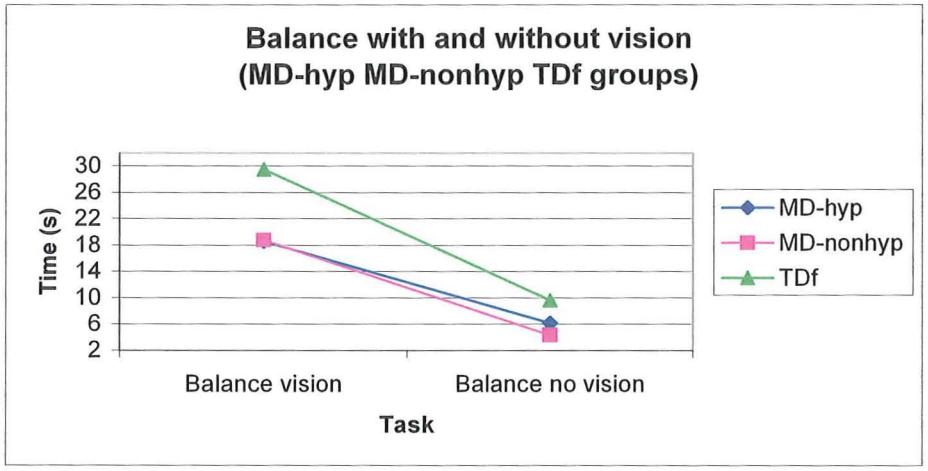


Figure 9.3b *Balance with and without vision for the DCDf, HFAf and TDF groups*



Examination of these data using two-way ANOVA repeated measures for TDf, MD-hyp, MD-nonhyp showed a main effect of vision, $F(1) = 103.7$, $p < .001$) and non significant vision-group interaction. Between subjects-group was significant, $F(2) = 5.5$, $p = .01$. Post hoc analyses were again dominated by the TDf proficiency (TDf-MD-hyp = .06 and TDf-MD-nonhyp = .01) but there was no significant difference between the clinical groups.

Failure to find support for either of the hypotheses tested was rather puzzling. However, one possible explanation stemmed from the wide individual subject variability that cut across all groups. For example, with vision, some children in all

five groups gained ceiling scores (TDf n = 7; DCDf n = 5; HFAf n = 3; MD-hyp n = 4 MD- nonhyp n = 7). However, the minimum individual score for DCDf was 2 seconds, for HFAf 4 seconds compared to the TDf individual minimum score of 27 seconds. Put another way, none of the TD children balanced less than 10 seconds whereas for the clinical groups the individual data showed that numbers who balanced less than 5 seconds in each group were DCDf 4; HFAf 2; MD-hyp 3; MD-nonhyp 3. This meant, of course, that these children had such low scores under normal conditions that there was no way of measuring the effect of removing vision i.e. there was a basement effect. When the individual data were examined for balance without vision condition the results were as follows: The TDf maximum score (seconds) was 20.3 with 4 children scoring over 10 seconds and 2 children balancing for </= 5 seconds. For the clinical groups the figures were DCDf maximum 18.2 (the only child in this group to score over 10 seconds and nine scored </= 5 seconds). The HFAf maximum was 8.3 and eight scored </= 5 seconds.

The maintenance of balance relies on a dynamic interaction between vision (especially utilising vertical and horizontal environmental cues), tactile/proprioception (through dermal receptors in the sole of the foot, muscle spindles and Golgi tendon receptors) in addition to vestibular calibration via semicircular canals. Clark and Watkins (1984) stressed the multidimensionality of static balance. Riach and Starkes (1993) noted that at around the age of 7 years (the lower cut off point for inclusion in the present study) children demonstrate mature balance control similar to adults. Although the measurement of the more complex aspects of balance control require sophisticated equipment, there are aspects that can be observed qualitatively by a trained observer. This training in observation is a core clinical skill for physiotherapy practice. One of the advantages of the present study, therefore, was the researcher's experience, and familiarity in observing a wide variety of children with balance problems. All of these require observation of gross and fine postural adjustment, and often involvement in decisions regarding the provision of orthoses, surgery aimed at releasing or fixing key points of control. The children in the study showed a range of problems and attempts to solve these. Most obvious was the muscle activity around the ankle but also in the small muscles of the foot. Some children showed continuous active adjustment in these muscles whereas others remained static with no visible muscle action. In the vision-occluded condition the activity was increased but in addition action often spread to the upper body with

trunk flexion and side flexion coupled with shifting on the supporting foot. Other children just swayed slightly and quickly put their foot down for support. These different observations of muscle action especially around the ankle support the findings in more sophisticated laboratory-based studies that may indicate that different strategies are used to maintain balance and children may adopt a variety of strategies to constrain their degrees of freedom (Winter, 1995). Geuze (2003), mentioned earlier, selected a sub-group of children aged 6-11 years with 'DCD' and poor static balance. He reported that all children found balance on one leg and with eyes closed harder and that children with DCD with balance problems showed more active control (reflected in centre of pressure displacement on force platform scores) than comparison children and especially in the one-leg stance eyes closed condition and displacement in the lateral as opposed to anteroposterior direction indicating a stance-direction interaction. However, he found no main effect of vision/no vision between the two groups and did not, of course include children from any other diagnostic category. His results, using EMG measures of muscle activity for anterior and lateral ankle control and knee/ hip stabilisation, indicated that less ankle activity occurs with age, as balance matures. In the DCD group with balance problems there was significantly greater/more frequent episodes of co-contraction of lower and upper leg muscles. He concludes that there is individual variability and that only a sub-group of children with DCD and balance problems are unable to perform in the normal range and rely on more co-activation of key ankle and hip muscles. His third experiment examined the effect of perturbation (see also Fawcett & Nicolson, 1999) once balance on two legs was in place. Groups reacted quite similarly but DCD children with balance problems seemed less efficient at regaining disturbed balance following perturbations.

In summary, the present hypothesis that all children would have more difficulty balancing with eyes closed was, not surprisingly, supported but the proposals regarding either pair of clinical groups was not. The fact that such large individual variability within each group was present may mask potentially interesting features in the group profiles. On the other hand, it may just reflect that any differences occur within rather than across groups defined on diagnostic measures.

The effect of a secondary task: peg-turning or balancing while counting

The previous section compared the five groups of children on simple tasks under different perceptual conditions. This section utilises a dual-task paradigm to examine the effect of a concurrent cognitive task on the simple peg-turning or balancing items that the children had already completed and were reported above. Dual-task methodology involves a primary and secondary task carried out simultaneously e.g. balancing combined with auditory reaction time in children or adults (Nicolson & Fawcett, 1990). Several research studies have used this method to examine attentional demands and automaticity of postural tasks and the effect of cognitive tasks on motor performance (Pellecchia, 2003; Nicolson & Fawcett, 1990). Regulation of posture is not initially automatic (a young child is unsteady) but gains automaticity through dynamic 'tuning' between the child and his environment through active experience and practice. Nicholson and Fawcett (1990) suggested that conscious resources were involved in monitoring balance which impacted a secondary cognitive task and concluded that motor function (which they relate to the cerebellum) was poorly automomized in children with developmental conditions such as dyslexia.

Peg-turning, with and without counting.

Manual dexterity is an area commonly reported as difficult for children in the clinical groups chosen for the present study. Its importance is reflected in its inclusion as one of the main components of the M-ABC and other assessment batteries. Lack of manual dexterity compromises many everyday tasks such as fastening buttons, using cutlery or differentiating coins in one's pocket. For most of us, performance of these well learned tasks takes up little attention and we can carry on a conversation at the same time. Whether this is true of children with movement difficulties is often discussed but rarely investigated. Turning over a small peg within a narrow hole requires a fair degree of finger dexterity and visual perceptual accuracy. In this component of the study, we predicted that both the DCD and HFA groups would be relatively more affected by the secondary task than the TD group. A weaker prediction was that the HFA children would be even more affected than the DCD, because some would be more distracted by the counting element especially those children fascinated by number sequences. Since hypermobility is a 'physical' symptom, which often affects the fingers and proximal shoulder stability, we predicted it would make the peg-turning task more difficult than for TD children but

could see no reason why the addition of the counting task would have a differential effect.

Observation: Turning small pegs and turning small pegs + counting

Many children had difficulty with in-hand manipulation (turning the small pegs) but none was unable to complete the task once started. Children varied in their counting ability but generally a level was chosen which ensured a challenge. However, some children became very muddled when counting although they had shown an ability to sequence numbers during practice during part one of the assessment. Counting was recorded and again the children varied, with some speaking in almost inaudible whispers and others who obviously enjoyed the experience and asked to listen to their recording and even added more comments. When counting many children coupled the counting with peg placing but for some the two aspects of the task were carried out unrelated. For some the first number e.g. 3 was spoken as they placed the third peg and they delayed saying six until they were ready to place peg number 6. Clearly, detailed analysis of this aspect of task performance would clarify the findings. However, there is no space to deal with it here .

Results

Figures 9.4a and 9.4b show the results of the peg turn and counting experiment for the TDf children and the clinical groups.

Figure 9.4a *Results of the dual-task peg turn and counting experiment for the TDf, DCDf and HFAf groups*

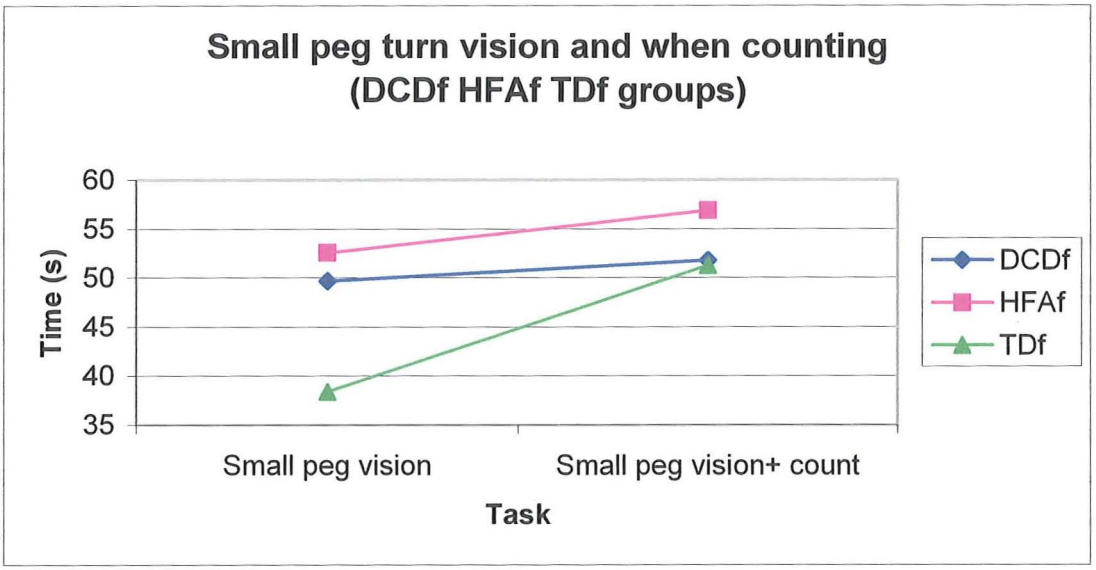
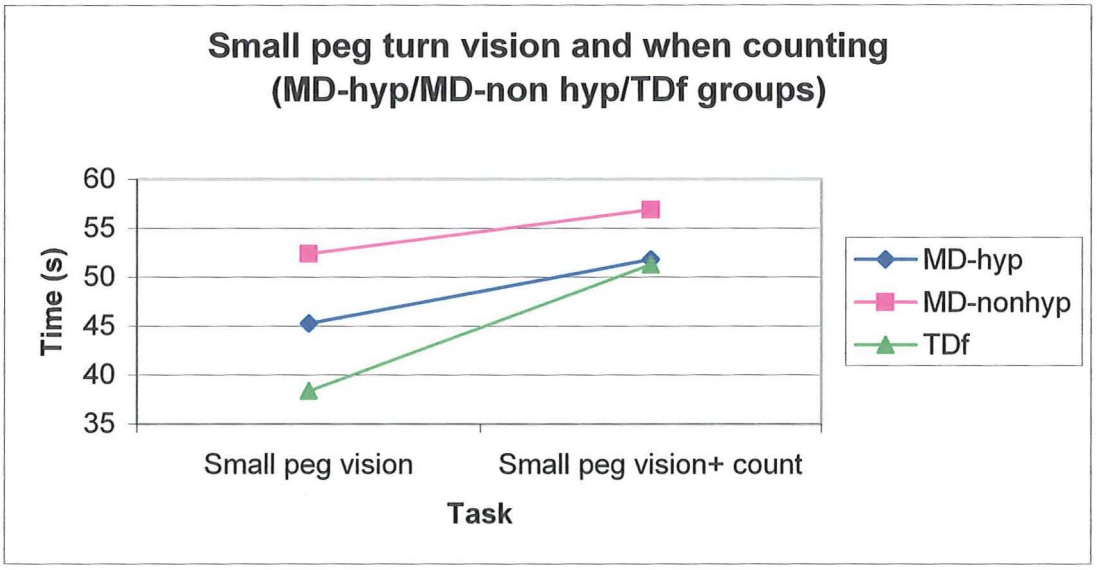


Figure 9.4b *Results of the dual-task peg turn and counting experiment for the TDf, MD-hyp and MD-nonhyp groups*



Results of the TDf DCDf and HFAf groups using two way ANOVA repeated measures showed that neither the main effect of group, $F(2) = 2.3$, $p = .1$, nor effect of task (no counting/counting), $F(1) = 2.3$, $p = .4$ was significant. Interaction between group and experimental manipulation was not significant ($p = .4$). The results for the MD-hyp and MD-nonhyp revealed a main effect of no counting/counting task, $F(1) = 4.4$, $p = .04$ but no other significant main effects or interactions.

As in the previous experiment there was wide variability of individual scores especially in the counting condition where scores were almost identical across all five groups (both Mean and SD). Interestingly, although the effect was not statistically significant, the addition of a cognitive task slowed the peg turning in the TD group most whereas the clinical group's scores slowed very little. This may imply that the clinical groups were already so slow on peg-turning (possibly already overloaded cognitively in executing the motor actions) that the cognitive task presented comparatively minor additional challenge and demand for switching attention.

Preferred leg balance and preferred leg balance+ counting.

For the balancing task, we were able to build upon the findings of Geuze, Nicholson and others, which suggested that any child with a movement problem would find the addition of a secondary task more difficult to cope with than a TD child. In addition, we predicted that the HFA children would be even more affected than the DCD for the same reasons as we felt they might be more distracted in the peg-turning task.

Observation

The children produced a variety of strategies to cope with the addition of the counting task to the baseline single leg balance experiment. For some of the typically developing children who achieved stable balance for over 30 seconds and who proudly mentioned their maths expertise the task presented no difficulty in spite of electing to count in 4s or 6s compared to other children who could barely manage to count in twos. Others who balanced equally well in the simple condition were obviously challenged and balance became observably less stable or counting became very hesitant.

Results

Results of dual-task balance counting experiment for the TDF, DCDf and HFAf groups are shown in Figure 9.5a and for the TDf MD-hyp and MD-nonhyp in Figure 9.5b. Two-way ANOVA repeated measures on the data in Figure 9.5a showed that neither the main effect of group, $F(2) = .7$, $p = .5$, nor effect of task (balance no counting /balance counting), $F(1) = .9$, $p = .4$ was significant. Interaction between group and experimental manipulation was, however, significant ($p < .01$).

Post hoc comparisons revealed significant difference between TDf-DCDf and TDf-HFAf groups (in each instance $p = .002$) but there was no difference between the two clinical groups. ($p = .9$).

The results for the TDF, MD-hyp and MD-nonhyp showed no main effects (min $F = .8$) but between subjects group effect was significant, $F(2) = 10.2$, $p < .01$. Pair-wise comparisons (with Scheffé correction as previously) gave a significant effect for the TDf and clinical groups (MD-hyp $p = .01$; MD-nonhyp $p < .01$). The MD-hyp and MD- nonhyp post hoc analyses were non-significant.

Figure 9.5a *Result of the dual-task counting/balance experiment for the TDf, DCDf and HFAf groups*

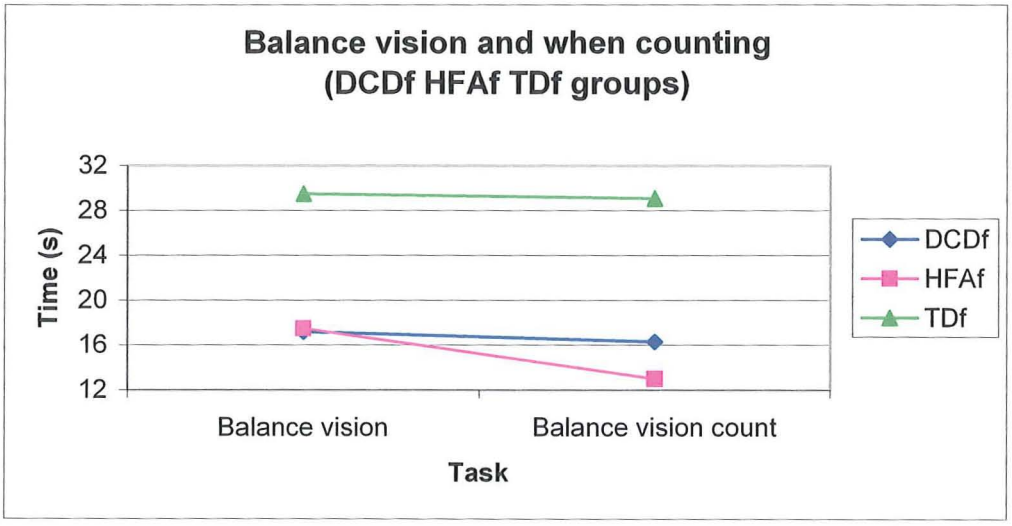
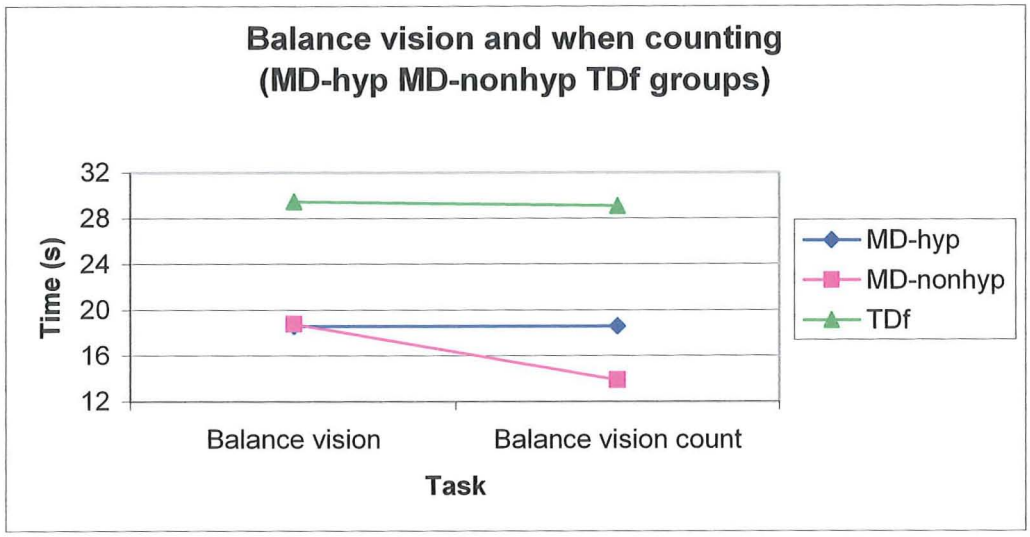


Figure 9.5b *Result of the dual-task counting/balance experiment for the MD-hyp and MD-nonhyp and TDf groups*



In summary, the dual task paradigm seemed to offer a promising way of examining subtle aspects of motor dysfunction in children. The addition of a secondary task once again revealed differences between the TDf and clinical groups but failed to reveal significant differences between the clinical groups. The small numbers in the experiments and the subject variability that cut across groups may confound results and could account for the lack of significant results. Qualitative observations suggested that there were some trends, such as timing of muscle actions and strategies adopted under different conditions (e.g., choosing pegs by size or rank order) which would be valuable to examine in greater detail with larger groups, more than one observer and more sophisticated measurement tools.

9.3.3 Conclusion

The first part of this study addressed the question of whether DCD exists as a separate syndrome, which can be clearly differentiated from other syndromes with clumsiness as a feature. Children who were thought to have DCD were compared to two other clinical groups of children, one bearing a label HFA, the other BJHS and to an age-matched comparison group. The results showed, that although the numbers of children might be small, so-called 'pure' cases do exist. Several features of the design of this study strengthened this claim. Great care was taken to reduce bias as far as possible. The participants were similar in that all were boys with IQ > 80 attending mainstream schools in the London area. The researcher tested the children

'blind' and only the child's name and age were revealed prior to assessment, i.e. the researcher knew neither the referral source nor any details of the child's diagnosis or developmental history. Two-step identification is often advised (Wright & Sugden, 1996b) and in this study attempt was made not only to accept children who had gone through an appropriate diagnostic assessment but also to confirm/reject the diagnisis given by assessing all children on tests that were appropriate for each category of child in the study. In other words, there was no prior presumption of a child's diagnosis and thus every child, including those referred as 'typically developing' completed screening tests for DCD, HFA and BJHS. This was an essential process in helping to ensure that for every child a definite answer might be given as to whether they had or did not have a 'pure' motor problem, were on the Autistic spectrum, or had joint hypermobility. Data related to anthropomorphical, medical, developmental and educational issues additionally helped to identify any factors that might bias the results.

That a few children presented with just a movement problem supports the long held view that a 'clumsy' child syndrome exists. However in the course of weeding out the pure cases the study supported the observation of others of how often developmental conditions present with many faces and not infrequently the presentation is so complex that a child cannot be fitted precisely into any one diagnostic category. In order to help ensure clearly defined groups of children in the second part of the study the cohort was somewhat reduced to include only those children who had a movement difficulty and met stringent criteria for DCD, HFA or typical development. When no such objective measures are included the results represent an ill-defined group on which to base any findings.

The second part of the chapter then turned to the question of whether the movement difficulties experienced by children with DCD or HFA, were the same or different. An examination of the profiles of performance on selected standardised tests was followed by analysis of performance on a series of novel experimental tasks, in which an increase in perceptual complexity, the removal of visual feedback and the addition of a secondary task were systematically employed. The theoretical basis for choosing this approach lay in the dynamic systems model of body- environment interaction (Thelen et al., 1987). Neither the perceptual, motor nor cognitive 'systems' function independently. The acquisition of smooth, automatic skilled

action arises through fine-tuning of all body systems within the constraints of the unique configuration of the individual, the demands of the task and the environment. The allocation of attention varies with the demands of the task and also varies with the competence of the individual. As an example we might walk confidently along a dry footpath but if the same path were wet and slippery or icy we would slow down and take greater care on how and where to place our feet. Similarly although we might succeed in aiming a ball through a netball goal we may be unable to thread a needle. As demand alters we adapt our response and in so doing our competence or lack of it may be highlighted. All our hypotheses were based on the idea that manipulation of aspects of this complex system might reveal differences between the clinical groups.

In spite of rigorous attention to detail very little support was found for the idea that 'clumsiness' in a developmental condition such as DCD differs from that of HFA. Whether this could be generalised to other conditions remains to be seen. From all of the comparisons conducted, only one statistically significant difference emerged between the DCD and HFA children and none between the hypermobile and non-hypermobile. The difference in balls skills replicated the finding by Green et al. (2002) who showed that a group of children with Asperger's, diagnosed on the same measures as the HFA group in the present study were also poor on the ball skills component of the M-ABC.

Chapter 10
Conclusions

Under the umbrella of neurodevelopmental disorders lies an ever expanding list of conditions: attention deficits (ADD, ADHD) autistic spectrum disorders (AS, HFA, PDD-NOS) movement disorders (Developmental Dyspraxia, Dysgraphia, DCD), reading disorder/dyslexia (RD), non-verbal learning disability (NVLD), speech and language impairment (SLI) and complex combined disorders (DAMP), to name but a few. As anyone familiar with even one of these terms will agree, DCD, the focus of this thesis, is not alone as an entity whose very existence has been questioned by many. Over the last fifty years, controversy over terminology within the broad category of neurodevelopmental disorders has raged on without solution. At the heart of this debate, of course, has been how we deal with the idea that damage to the Central Nervous System seems likely to be involved but manifests itself in so many different ways. For some revealing thoughts on this thorny issue, there is no better place to turn than the literature on minimal brain dysfunction, a term still applied to children with DCD in parts of Australia. In a book entitled "Minimal Brain Dysfunction: Fact of Fiction (Kalverboer, Praag & Mendlewicz, 1978), Praag in his introduction, states: *" Is minimal brain dysfunction (MBD) fact or fiction? Of course it is a fact. Who would want to refute that the functional integrity of the brain can be slightly disturbed. Is MBD as a diagnostic entity fact of fiction? This is a controversial question, which is not surprising because MBD is an ill-defined notion, vague and therefore, multi-interpretable" (p. 2).* After discussing both hyperactivity and delayed motor competence as components of the syndrome, Kalverboer (ibid) continues: *"..hardly any concept in the clinical nomenclature has occasioned so much controversy and confusion".* Put in a nutshell, the concept of **minimal** brain dysfunction resulted in **maximal** confusion.

The demise of terms encompassing any reference to the brain and its integrity, however, did not lead to immediate clarity. Rather than one box, we then found a proliferation of boxes, each in itself spawning controversy. For example, in 1987, we find a paper by Ayres and colleagues (Ayres et al., 1987) entitled "Is dyspraxia a unitary function?"; Taylor and McKinley (1979) posed the question " What type of thing is being clumsy?" Badawi (1998) asks "What constitutes cerebral palsy?;

Stanovich (1994) heads his annotation with the question: "Does dyslexa exist?"; Whitmore, & Bax, (1999) challenge by asking: "What do we mean by SLD?" and most recently Mayes et al (2001) ponder on yet another diagnosis with "Does DSM-IV Aspergers's disorder exist?" Bax (1999), referring to DCD as one of a group of specific learning difficulties/neurodevelopmental disorders, notes that *"terminology is a morass"* and that in consequence epidemiology and diagnosis are extremely difficult. In yet another attempt to solve the problem, of course, the wheel has now gone full circle and terms such as atypical brain damage (Kaplan et al., 1998) have been introduced to encompass the entire range of developmental disorders.

If terms are confused the entire infrastructure for communication both within and across the research & clinical communities is compromised. The general aim of this thesis, therefore, was to contribute to our understanding of **movement** disorders in children by examining the concept of DCD, theoretically and empirically. The two questions addressed in the thesis were (i) Does DCD exists as a discreet syndrome and (ii) if such a syndrome is identified, is the movement difficulty that defines it the same or different to movement difficulty present in other disorders. To a certain extent, these are not two separate questions but two facets of the same question. However, there are aspects of each which can de dealt with independently, as the empirical studies reported in this thesis illustrate.

Begining with the question of existence, three facts now seem incontestable. Both the literature review and the empirical studies undertaken in this thesis show that a) whatever label is adopted to describe it, abnormal clumsiness of movement in bright children, does exist and continues to be of increasing concern to professionals in health and education, to parents and most of all to the children themselves, b) there are children for whom the movement difficulty is either entirely isolated or is clearly primary (i.e. of over-riding importance) when compared with any other accompanying difficulty, and c) that compared to reading, speaking or behavioural problems abnormal clumsiness is, as Gillberg (2003) mentions, still "the black sheep" in the field of investigative research.

The first two empirical studies in the thesis gathered together the perceptions of different professionals working with children with movement difficulties and of parents. These two studies, along with the case histories, supported the idea that a

motor-based syndrome, nowadays subsumed under the title DCD, does indeed exist but that awareness, knowledge, and use of labels is not uniform. As noted above, such lack of clarity compromises communication between clients and all levels of service provider, has a knock on effect on identification of the disorder, and ultimately threatens service delivery and effective intervention. Taken together, studies 1 and 2 revealed that the perceptions of UK education and health professionals also tend to reflect a narrow uni-professional interpretation of DCD, and that those who do know the term DCD often fail to observe official criteria, using the term loosely to apply to any child referred to their clinic because of a mild to moderate movement difficulty. Parents' views also indicated that the label given to their child affects the pathway from initial concern to effective intervention.

Qualitative data collected in Studies 1, 2 and 4 raised another important point about labelling related to change over time. A label of SLI may be appropriate prior to school entry but a diagnosis such as dyslexia or dysgraphia would only become applicable later and further diagnoses may ensue. In adulthood, for example psychiatric disorders including personality and mood disorders are frequently reported. Several case studies showed that as the child gets older not only do their problems change but the impact on daily function alters. A proportion of children demonstrate pure movement difficulty initially but as time passes the number and extent of associated problems increase. With the changing presentation of symptoms children are frequently moved in and out of sequential diagnostic 'boxes' like pawns on a chess board. The studies also showed that professionals tended to focus for too long on symptoms central to their professional expertise thus extending the time before problems in other domains were recognised. Although the case studies did show both positive as well as negative aspects of labels, uncertainty and delay in applying a label, clearly had emotional and practical effects on family life. The way forward is through a 'joined-up' model that encourages better communication through closer intra and inter- agency team working.

All five studies, that included more than a hundred and fifty children in total, provided evidence for the existence of 'pure' cases of movement difficulty with no, or at least relatively minor, associated problems in other domains. In these cases, the movement difficulty was seen as primary and other aspects appeared to be the consequence of the movement difficulties. However, far more common was overlap,

where the child presented with symptoms of movement difficulty accompanied by problems in other domains. Thus, a child with DCD might also fit the AS, ADHD, SLI, or Dyslexia box, or perhaps even fit into several diagnostic categories. By selecting objective assessment tools across domains of function, Studies 3 and 5 showed that it was possible to identify and separate those children with a primary movement difficulty from those with movement problems secondary to, or associated with, other diagnoses. A significant proportion of boys in Study 5 referred with 'pure' DCD or BJHS in fact ended up as unquestionably more appropriately bearing a label of HFA/AS. A major issue, which needs to be considered in this field, is the trend toward recognition of the dimensionality of many conditions. For example, we have a continuum implied in autism spectrum disorder and in other conditions such as foetal alcohol syndrome which has recently been renamed foetal alcohol spectrum disorder. This allows for recognition of phenotypic variability and reduces the splintering of diagnostic categories into too many pieces. The literature on DCD makes little mention of a continuum of movement disorder, except in the context of the dividing line between cerebral palsy and DCD.

An important aspect of the overlap between conditions is its dependence on where one chooses to draw the boundaries. Convergence of one syndrome on another, such as Asperger and non-verbal learning difficulty (Klin et al 1995) or CP and DCD (Badawi 1998) emphasises two other important points: the value of medical screening and the need for breadth in assessment. Figures 9.2a and 9.2b in Study 5 show just how the picture many be manipulated by choice of cut-off points. The need for medical screening, was supported in all five studies, as essential for identification of DCD as a discreet entity since children in each study failed to meet Criterion C as currently specified in DSM-IV. Most research purporting to focus on DCD fails to check for the presence of a medical condition. Here again the picture reveals inconsistency in the levels of investigation. Gross neurological hard signs or muscle weakness may clearly identify an alternative diagnosis on one level, but are rarely assessed. Moreoever, as current technology becomes ever more sophisticated, brain scans, biochemical, physiological and genetic studies, are likely to open up the aetiological black box and impact on diagnostic issues in a way never previously imagined. Dynamic systems theory emphasises environmental interaction and this includes the internal environment at a sub-cellular level that shapes the eventual phenotype. Resultant atypical development may also emanate from cells other than

brain cells. Finally, breadth of assessment relates to identification and the exclusion of confounding problems outside the motor domain. Breadth of assessment is vital if the core motor impairment is to be better understood.

In summary, although progress has been made there continues to be confusion regarding the precise nature of the disorder, DCD. Even if, at present, we decide to proceed as if it existed, the question of how one defines DCD remains: in terms of aetiology, underlying systems and processes affected, or along functional lines at the level of the phenotype? There are many different ways to cut the terminological cake: on the one hand we have a plethora of 'separate' disorders in DSM and ICD constrained by their specific categorical labels and on the other there is the recent attempt toward convergence of conditions under more global terms such as DAMP and ABD. Yet another solution can be found in the move away from the diagnostic medical model of classification of impairment, disability and handicap (WHO, 1980) toward emphasis on function in the International Classification of Function (WHO 2001; 2002). Here, the emphasis is on health rather then disease and not only is it coded on 'body function', 'activity' and 'participation' but also along dimensions which reflect the environment, social impact and severity. Whatever system one uses, however, it is important to be aware that terminology and its employment is dynamic as opposed to being set in pillars of stone and the exploration of new ideas, which may at times seem retrograde, might in the end be productive. Understanding DCD, as a discreet syndrome- or not - depends upon keeping abreast of the changes in emphasis but, for the moment, we must continue to interpret and apply officially recognised criteria for DCD unequivocally and no longer rely on non-standard observation of a child's movement. An encouraging way to drive forward along this route is through meetings such as those in London, Ontario and Leeds, UK which led to clearly formulated consensus statements (Fox, & Polatajko, 1994; Polatajko et al 1995; Leeds Consensus Statement, 2006). These have physically brought people together to discuss issues such as internationally acceptable standard criteria for DCD. Once researchers have blazed a trail, however, there remains much work to do within the classroom and clinic,where there still remains a worrying lack of understanding of DCD at both a theoretical and practical
level.

The second question addressed in this thesis was whether the movement difficulty in DCD is in any way different to that displayed in any other neurodevelopmental disorder. Whereas in the previous section the focus was on justifying a **syndrome,** labelled DCD, with movement as its defining symptom, the focus of the next section is on characterising the **movement symptoms** as displayed in DCD. The literature on DCD constantly refers to the heterogeneity within the motor domain in these children. In all five studies reported here, children who met criteria for DCD displayed a variety of motor strengths and weaknesses, with the objective data being well supported by parent and teacher observation. For example, the case studies confirmed that for some children manual dexterity was the main area of difficulty. For others it was balance that proved more challenging and for many the problems spanned every domain of motor function. Perhaps not surprisingly, the variable presentation of DCD has led some to suggest that the syndome encompasses systematic subtypes. Existing investigations provide some support for this notion with a degree of consistency emerging across studies but there is, to date, lack of evidence to suggest that this is either diagnostically or aetiologically linked. Also, in most studies, the description of the samples of children involved is sparse, ill-defined and leaves much to be desired. This makes comparison between studies almost impossible.

Study 3 in this thesis ventured into the difficult territory of subtyping via cluster analysis. This study showed that there were measurable differences in both the severity and in the area of movement difficulty affected. Four cluster groups were identified; one group was comparatively good at everything and a second group scored poorly across all variables. Contrasting with these 'flat' profiles two groups emerged with uneven profiles, reflecting dissociations between different elements of motor control. There were some similarities here between the findings of Jongmans (1993) and Hoare (1994), which might be worth pursuing. A novel aspect of this study was the inclusion of a group with known medical problems, which clustered together, alongside the DCD group with the poorest scores, which suggests support for the notion of an aetiological continuum of damage (Knobloch, & Pasamanick, 1959). Although there were aspects of Study 3 which might have been pursued, a lack of confidence in the usefulness of this approach led to its abandonment. In particular, the fact that the only real way to validate subtypes in any disease or

disorder is to show that each one reacts to a different type of intervention, seems a) premature in the field of DCD and b) unmanageable as part of a PhD project.

As mentioned previously, a sizeable minority of children displayed a primary motor problem, without significant dysfunction in non-motor domains, thus commensurate with a definite diagnosis of DCD. It is these 'unadulterated' cases that form the best group for in-depth comparison with other clinical groups, and it was children like this that formed the focus of Study 5. For comparison, practical considerations led to the inclusion of children with HFA/AS and BJHS. Once the comparison groups for this study had been decided, a major issue was the question of which measures might best produce a true and satisfactory profile of the childrens' movement strengths and weaknesses? Motor function is not a uni-dimentional construct and it may be measured in many different ways along a variety of dimensions. In some conditions there is a clear pathway that links the functional manifestation of the problem with underlying processing deficits e.g. one particular type of reading disorder is identified by tests that focus on difficulty in phonological processing. DCD has no clear equivalent where measures at different levels are directly linked. Similarly, at an aetiological level, unlike scarlet fever or measles it cannot be identified by the presence or absence of a bacterium or rash. At the present time even if one drew upon a hundred different measures one could not hope to capture every aspect of DCD. In addition, in practical terms there are limits to the time and costs available and the child's tolerance. For example, in Study 5, Ethics Committee directions constrained the length of assessment and insisted that rest periods be included. The final study therefore, contained a range of measures, each of which had their strengths and weaknesses. In the case of the M-ABC, for example, the fact that children who clearly meet criterion B for DCD do not 'fail' the test, suggests that there are important aspects of motor impairment, which are missed by the test. With regard to the experimental measures, their choice was not theoretically driven as should have been the case. Nevertheless, the study yielded a number of interesting findings, while at the same time raising a number of questions.

Negative findings are never easy to deal with but the fact that this study employed such a broad range of tests that were in some way or other "movement-related" at least strengthens the view that the movement difficulties experienced by children with different diagnoses are very similar. Starting with the standardised tests, we

consistently found that the typically developing children stood out as different from all other groups, which at least confirmed the validity of the tests, as reported in the various manuals. However, the results were not able to support a case for differentiation on the basis of diagnostic category. Only one small difference emerged on the ball skills component of the M-ABC. Children with HFA were significantly less proficient on the M-ABC ball skill component which supported the study by Green et al (2002). The recurrence of this finding renders it ever more intriguing. Many suggest that the problem may simply be due to lack of experience on the part of the AS/HFA children but a properly designed experimental study might tell a different story. On the standardised measures, the children with HFA also had the lowest motor scores, had been referred via a tertiary clinic (a very specialised route) and unlike the DCD group were all in the process of, or already receiving educational support in school, in spite of average IQ scores. This supports the observation of Gillberg (2003) that problems in the motor domain attract less attention compared to the impact of social, communication and attentional aspects. Although the restriction to boys in a narrow age range in the study was planned in order to further standardise the sample, it reduces the generalisability of these findings to other ages or to girls.

With regard to the experimental measures, once again there was no evidence to suggest that children from different diagnostic categories responded differently to the different manipulations. There are two possible explanations of this outcome. First, it might be argued that the choice of tests and their manipulation was misguided. Second, it might be argued that the choice of diagnostic groups was wrong and that children with dyslexia or ADHD would respond differently. However, I consider it more likely that specific features of clumsiness of movement may reflect across group variability rather than be diagnostically driven. Ultimately, if differences are found the next stage is to examine the validity of the difference in identification of differential response to intervention. An aim for the future must be to set up carefully controlled trials of specific intervention with larger samples of children whose movement difficulty has been clearly and similarly defined.

In summary, it is hoped that the thesis has added to the body of knowledge about DCD through the critical literature review and empirical studies in which qualitative and quantitative data on over 150 children were collected and analysed. The

conclusion is that DCD should at present be viewed as a discreet syndrome, which may occur in isolation or within a variety of diagnoses. Overall, however, the idea that motor aspects of DCD are quantifiably different from motor dysfunction in other conditions was not supported.

References

Abbie, M. (1974). Treatment of 'Minimal Brain Dysfunction' *Physiotherapy, 60,* 203-207.

Abbie, M. (1978). Physical treatment for clumsy children - not enough? *Physiotherapy, 64,* 198-203.

Adapted Physical Activity Quarterly (1994): *Developmental Coordination Disorder, 11*(2), 111-244. Special Issue

Adib, N., Davies, K., Grahame, R., Woo, P., & Murray, K. J. (2005). Joint hypermobility syndrome in childhood. A not so benign multisystem disorder? *Rheumatology, 44*(6), 744-750.

Adler, H. (1982). Children with problems in physical education in middle school need help and assistance after grade school. *Läkartidningen, 79,* 2878-80.

Aicardi, J. (1992) Diseases of the Nervous System in Childhood. Chapter 27 Deficits in attention, motor control and perception and other syndromes attributed to minimal brain dysfunction. [Monograph] *Clinics in Developmental Medicine, 115/118,* 1321-1337). Oxford: Mac Keith Press.

Alberman, E. & Stanley, F. (1984). Guidelines to the epidemiological approach. In F. Stanley & E. Alberman (Eds.), *The Epidemiology of the Cerebral Palsies.* [Monograph] *Clinics in Developmental Medicine, 87,* 172-183. Oxford: Spastics International Medical Publications.

Alderson, L., Peters, J. M., & Main, E. (2006). The GAITRite ® mat as a quantitative measure of dynamic walking balance in children with coodination problems. *Association of Paediatric Chartered Physiotherapists (APCP) Journal 118,* 4-10.

American Psychiatric Association (1952). *Diagnostic and Statistical Manual of Mental Disorders* (1st ed.). Washington, DC: American Psychiatric Association.

American Psychiatric Association (1975). *Diagnostic and Statistical Manual of Mental Disorders.* (2nd ed). Washington, DC: American Psychiatric Association.

American Psychiatric Association (1980). *Diagnostic and Statistical Manual of Mental Disorders.* (3rd ed.) Washington, DC: American Psychiatric Association.

American Psychiatric Association (1987). *Diagnostic and Statistical Manual of Mental Disorders* (3rd ed. rev.). Washington, DC: American Psychiatric Association.

American Psychiatric Association (1994). *Diagnostic and Statistical Manual of Mental Disorders: DSM-IV* (4th ed.). Washington, DC: American Psychiatric Association.

American Psychiatric Association (2000). *Diagnostic and Statistical Manual of Mental Disorders: DSM-IV* (4th ed.) *Text Revision (DSM-IV-TR).* Arlington VA: American Psychiatric Association.

Angold, A., Costello, E. J., & Erkanli, A. (1999). Comorbidity. *Journal of Child Psychology and Psychiatry, 40*, 57-87.

Annell, A. L. (1949). School problems in children of average or superior intelligence: preliminary report. *Journal of Mental Science, 95*, 901-909.

Arnheim, D. D., & Sinclair, W. A. (1975). *The clumsy child: a program of Motor Therapy.* St Louis: C.V. Mosby.

Ayres, A. J. (1965). Patterns of perceptual-motor dysfunction in children. A factor analytic study. *Perceptual and Motor Skills, 20*, 335-368.

Ayres, A. J. (1972). *Sensory Integration and Learning Disorders.* Los Angeles, CA: Western Psychological Services.

Ayres, A. J. (1980). *Southern California Sensory Integration Tests Manual: Revised.* Los Angeles, CA: Western Psychological Services.

Ayres, A. J. (1985). *Developmental Dyspraxia and Adult-Onset Apraxia.* Torrance, CA: Sensory Integration International.

Ayres, A. J. (1989). *Sensory Integration and Praxis Tests.* Los Angeles, CA: Western Psychological Services.

Ayres, A. J., Mailloux, Z. K., & Wendler, C. L. W. (1987). Developmental dyspraxia: Is it a unitary function? *Occupational Therapy Journal of Research, 7*, 93-110.

Ayyash, F. A., & Preece, P. M. (2003). Evidence-based treatment of motor co-ordination disorder. *Current Paediatrics, 13*, 360-364.

Badawi, N., Watson, L., Petterson, B., Blair, E., Slee, J., Haan, E., & Stanley, F (1998). What constitutes cerebral palsy? *Developmental Medicine and Child Neurology, 40*, 520-527.

Baker, J. (1981). A psycho-motor approach to the assessment and treatment of clumsy children. *Physiotherapy, 67*, 355-362.

Barnett, A. L., & Henderson, S. E. (1992). Some observations on the figure drawing of clumsy children. *British Journal of Educational Psychology, 62*, 341-355.

Barnett, A. L., & Henderson, S. E. (2005). Assessment of Handwriting in children with developmental coordination disorder. In D. Sugden & M. Chambers

(Eds.), *Developmental Coordination Disorder* (pp. 168-188) London: Whurr Publications.

Barnett, A. L., Mercuri, E., Rutherford, M., Haataja, L., Frisone, M. F., & Henderson, S. et al. (2002). Neurological and perceptual-motor outcome at 5-6 years of age in children with neonatal encephalopathy: relationship with neonatal brain MRI. *Neuropediatrics, 33,* 242-248.

Barnett, A., & Peters, J. M. (2004). Motor Proficiency Assessment Batteries. In D. Dewey, & Tupper, D. E. (Eds.), *Developmental Motor Disorders: A Neuropsychological Perspective* (pp. 66-109). New York: Guilford Press.

Barnett, L. A., Kooistra, L., & Henderson, S. E. (1998). Clumsiness as syndrome and symptom. *Human Movement Science, 17,* 435-444.

Barron, D., Cohen, B. A., Geraghty, M. T. Violand, V., & Rowe, P. C. (2002). Joint hypermobility is more common in children with chronic fatigue syndrome than in healthy controls. *Journal of Pediatrics, 141,* 421-425.

Bax, M. C. O. (1999). Specific leaning disorders/neurodevelopmental disorders. Editorial. *Developmental Medicine and Child Neurology, 41,* 147.

Bax, M., & Mackeith, R. (Eds.) (1963) *Minimal Cerebral Dysfunction, Little Club Clinics in Developmental Medicine* London: Heinemann.

Beery, K. E. (1989). *The Developmental Test of Visual-Motor Integration 3rd Revision (3R).* Cleveland, OH: Modern Curriculum Press

Beery, K. E. (1997). *The Beery-Buktenica Test of Visual-Motor Integration with supplemental Developmental Tests of Visual Perception and Motor Coordination.* Parippany, NJ: Modern Curriculum Press.

Beighton, P. H., Soskolne, L., & Solomon, C. L. (1973). Articular mobility in an African population. *Annals of the Rheumatic Diseases, 32,* 413-417.

Bishop, D. (2006). Developmental cognitive genetics: how psychology can inform genetics and vice versa. *Quarterly Journal of Experimental Psychology, 59,* 1153-1168.

Black's Medical Dictionary (2002). London: A & C Black.

Bobath, K., & Bobath, B. (1984). The neuro-developmental treatment. In: D. Scrutton (Ed.), *Management of motor disorders of children with cerebral palsy.* Oxford: Blackwell Scientific Publications Ltd.

Bos, A. F., Martin, A., Okken, A., & Prechtl, H. F. R. (1998). Quality of general movements in preterm infants with transient periventricular echo-densities. *Acta Paediatrica, 87,* 328-335.

Bouffard, M., Watkinson, E. J., & Thompson, L. P. (1996). A test of the Activity Deficit Hypothesis with children with movement difficulties. *Adapted Physical Activity Quarterly, 13,* 61-73.

British Heart Foundation (2000). *Coronary Heart Disease Statistics.* London: BHF.

Brown, K. (1996). Will he grow out of it? (Editorial). *Developmental Medicine and Child Neurology, 38,* 189-190

Brown, K. J., Tarek, O., & O'Regan, M. (1997). Brain development and the development of tone and movement. In K. J. Connolly & H. Forssberg (Eds.), *Neurophysiology and Neuropsychology of Motor Development.* [Monograph] *Clinics in Developmental Medicine, 143/144,* 378. London: Mac Keith Press.

Bruininks, R. H. (1978). *The Bruininks-Oseretsky Test of Motor Proficiency.* Circle Pines, MN: American Guidance Service.

Bulbena, A., Duro, J. C., Porta, M., Faus, S., Vallescar, R., & Martin-Santos, R. (1992). Clinical assessment of hypermobility of joints: assembling criteria. *The Journal of Rheumatology, 19,* 115-121.

Burnham, J. (personal communication, 2005). *Before Accident Proneness.* Unpublished Paper. Ohio State University.

Campbell, J. C. (2005). Diagnostic assessment of Asperger's disorder: A review of five third-party rating scales. *Journal of Autism and Developmental Disorders 35,* 25-35.

Campbell, P. N., Smith, A. D., & Peters, T. J. (2005). *Biochemistry Illustrated: Biochemistry and Molecular Biology in the Post-genomic Era* (5th ed.) Edinburgh: Elsevier.

Cans, C. (2000). Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. *Developmental Medicine and Child Neurology, 42,* 816-824.

Cantell, M. H., Smyth, M. M., & Ahonen, T. P. (1994). Clumsiness in adolescence: Educational, motor, and social outcomes of motor delay detected at five years. Special Issue of *Adapted Physical Activity Quarterly, 11,* 115-129.

Cantell, M., & Kooistra, L. (2002). Long-term outcomes of developmental coordination disorder. In: S. A. Cermak & D. Larkin (Eds.), *Developmental Coordination Disorder* (pp. 23-38). Albany, NY: Delmar Thomson Learning.

Cavill, N., Biddle, S., & Sallis, J. (2001). Health enhancing physical activity for young people: statement of the United Kingdom Expert Consensus Conference. *Pediatric Exercise Science, 13,* 12-25.

Cermak, S. A., & Larkin, D. (Eds.) (2002). *Developmental Coordination Disorder* Albany, NY: Delmar Thomson Learning.

Cermak, S. A., Ward, E. A., & Ward, L. M. (1986). The relationship between articulation disorders and motor coordination in children. *American Journal of Occupational Therapy, 40,* 546-50.

Chartered Society of Physiotherapy (2005). *The Health Professions Council (HPC) Standards for Continuing Professional Development (CPD)* [Information Paper CPD39]. London: CSP.

Chartered Society of Physiotherapy (CSP) (1995). *Standards of Practice for Paediatric Physiotherapy.* London: Association of Paediatric Chartered Physiotherapists.

Chartered Society of Physiotherapy (CSP) (2000). *Standards of physiotherapy practice (3rd ed.)* London: Chartered Society of Physiotherapy.

Chartered Society of Physiotherapy (CSP) and College of Occupational Therapy (COT) (1999). *The Garner Report: Scoping the information management needs in occupational therapy and physiotherapy.* London: Chartered Society of Physiotherapy.

Clark, J. E., & Watkins, D. L. (1984). Static balance in young children. *Child Development, 55,* 854-857.

Clements, S. G., & Peters, J. E. (1962). Minimal brain dysfunction in the school-age child. *Archives of General Psychiatry, 6,* 185-197.

Cole, T. J., Bellizzi, M. C., Flegal, K. M., & Dietz, W. H (2000). Establishing a standard definition for child overweight and obesity worldwide: international survey. *British Medical Journal, 320,* 1240.

Coleman, R., Piek, J., & Livesey, D. J.(2001). A longitudinal study of motor ability and kinaesthetic acuity in young children at risk of developmental coodination disorder. *Human Movement Science, 20,* 92-110.

Collier, J., & Adie, W. (1922). Diseases of the Nervous System. in *Price's Textbook of Medicine* (1st ed) (pp. 1332-1711)

Collins Shorter Dictionary and Thesaurus (1995). Glasgow: HarperCollins.

Concise Oxford English Dictionary (2004). 11th Edition. Oxford, UK: Oxford University Press.

Cousins, M., & Smyth, M. M. (2003). Developmental coordination impairments in adulthood. *Human Movement Science, 22,* 433-59.

Cratty, B. J. (1994). *Clumsy child syndromes: description, evaluation and remediation.* London: Harwood Academic Publishers.

Davies, P. A. (1994). Speaking the same language. Editorial. *Developmental Medicine and Child Neurology, 36,* 189-190.

DCD Research UK (2004). *A Conference on Research into Developmental Coordination Disorder in the UK.* Oxford, UK.

DCD Research UK-II (2006). *A Conference on Research into Developmental Coordination Disorder.* Oxford, UK.

DCD V (2003). *5th International Conference on Developmental Coordination Disorder*. Banff, Canada.

DCD VI (2005). *6th International Conference on Developmental Coordination Disorder*. Trieste, Italy.

Denckla, M. (1978). Minimal brain dysfunction. In J. S. Chall, & A. F. Mirsky (Eds.), *Education and the Brain* (pp. 223-268). Chicago: University of Chicago Press.

Department for Education (DoE) (1988). *Education Reform Act*. London: HMSO.

Department for Education (DoE) (1994). *Code of practice on the identification and assessment of Special Educational Needs*. London: HMSO.

Department for Education and Skills (DfES) (2001) *SEN Toolkit* (DfES 558), Annesley: DfES.

Department of Education and Science (DES) (1981). *Education Act - 1981*. HMSO. London.

Department of Health (DoH) (1999). *Electronic patient records move a step closer*. [Press Release]. London: Department of Health.

Department of Health (DoH) (2004). *At least five a week: Evidence on the impact of physical activity and its relationship to health*. London: Department of Health.

Dewey, D., & Kaplan, B. (1992). Subtyping of developmental motor deficits. *Journal of Clinical and Experimental Neuropsychology, 14*, 50.

Dewey, D., & Kaplan, B. (1994). Subtyping of developmental motor deficits. *Developmental Neuropsychology, 19*, 265-284.

Dewey, D., & Tupper, D. E. (Eds.) (2004). *Developmental Motor Disorders: A Neuropsychological Perspective*. New York: Guilford Press.

Dewey, D., Kaplan, B., Crawford, S. G. et al. (2002). Developmental coordination disorder: associated problems in attention, learning and psychosocial adjustment. *Human Movement Science, 21,* 905-918.

Dorland's Illustrated Medical Dictionary (24th Ed.) (1965). Philadelphia: W. B. Saunders Co.

Duel, R. K., & Doar, B. P. (1992). Developmental manual dyspraxia: a lesson in mind and brain. *Journal of Child Neurology, 7,* 99-103.

Dunford, C., & Richards, S. (2003). *Doubly disadvantaged.* Report of a survey on waiting lists and waiting times for occupational therapy services for children with developmental coordination disorder. *The College of Occupational Therapists and National Association of Paediatric Occupational Therapists.*

Dunford, C., Street, E., & O'Connell, H. (2004). Are referrals to occupational therapy for developmental coordination disorder appropriate? *Archives of Disease in Childhood, 89*, 142-147.

Dunn, L., Dunn, L., Wetton, C., & Burley, J. (1997). *British Picture Vocabulary Scale* (2nd ed.) (BPVS II) Windsor : National Foundation for Educational Research (NFER-NELSON).

Duprè, E., & Merklen, P. (1909). L'Insuffisance pyramidale physiologique de la première enfance et le syndrome de débilité motrice. Revue. *Neurologique, 17*, 1073-1074.

Dyspraxia Foundation (1997). Developmental Dyspraxia Explained. [Information Leaflet]. Hitchin: Dyspraxia Foundation.

Editorial (1919). The human factor in accident occurrence *Engineering and Industrial Management, 9*, 261-262.

Editorial (1962). Clumsy Children. *British Medical Journal,* 1665-1666.

Edwards, R. H. T. (2002). Forword (p. xvii) in V. R. Preedy & T. J. Peters (Eds.), *Skeletal Muscle Pathology, Diagnosis and Management of Disease*. Oxford: GPG Books.

Ehlers, S., Gillberg, C., & Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders, 29*(2), 129-141.

Einspieler, C., Prechtl, H. F. R., Bos, A. F., Ferrari, F., & Cioni, G. (2005). Prechtl's Method on the Qualitative Assessment of General Movements in Preterm, Term and Young Infants. *Clinics in Developmental Medicine,* 167, 1-91. Cambridge: Mac Keith Press.

Emery, A. E. H., & Emery, M. L. H. (1995). *The History of a Genetic Disease. Duchenne Muscualr Dystrophy or Meryon's Disease.* London: The Royal Society of Medicine Press.

Fawcett, A .J. & Nicolson, R. I. (1999). Performance of dyslexic children on cerebellar and cognitive tests. *Journal of Motor Behavior, 31*, 68-78.

Fawcett, A. J., & Nicolson, R. I. (1992). Automisation deficits in balance for dyslexic children. *Perceptual and Motor Skills, 75,* 507-529.

Fawcett, A. J., & Nicolson, R. I. (1995). Persistent deficits in motor skill of children with dyslexia. *Journal of Motor Behavior, 27*, 235-240.

Fawcett, A. J., Nicolson, R. I., & Dean, P. (1996). Impaired performance of children with dyslexia on a range of cerebellar tasks. *Annals of Dyslexia, 46*, 259-283.

Ferrari, J., Parslow, C., Lim, E., & Hayward, A. (2005). Joint hypermobility: the use of a new assessment tool to measure lower limb hypermobility. *Clinical and Experimental Rheumatology, 23*, 413-420.

Fisher, A., Murray, E. A., & Bundy, A. C. (Eds.) (1991). *Sensory Integration, Theory and Practice*. Philadelphia: F. A. Davis Company.

Fog, E., & Fog, M. (1963). Cerebral inhibition examined by associated movements. In M. Bax & R. MacKeith (Eds.), *Minimal Cerebral Dysfunction. Clinics in Developmental Medicine,* No 10. London: Spastics International Medical Publications with Heinemann Medical Books Ltd.

Ford, F. R. (1959). *Diseases of the Nervous System in Infancy, Childhood and Adolescence*. Springfield: Thomas.

Forseth, A. K., & Sigmundsson, H. (2003). Static balance in children with hand-eye co-ordination problems *Child: Care, Health and Development, 29*, 569-579.

Fox, A. M., & Polatajko, H. J. (Eds.) (1994). *The 'London Consensus': from Children and Clumsiness: An International Consensus Meeting,* London Ontario. Canada. 11th-14th October.

Frith, U. (1991). *Autism and Asperger's syndrome.* Cambridge: Cambridge University Press.

Frostig, M. (1971). Cerebral minimal syndrome: psychological and educational treatment. *XIII International Congress of Pediatrics, Vienna, Vol. 3,* (p. 11-28), *Neurology and Psychiatry,* Vienna: Weiner Medizinische Akademie.

Gadow, K. D., Nolan, E. E., Sprafkin, J., & Schwartz, J (2002). Tics and psychiatric comorbidity in children and adolescents. *Developmental Medicine and Child Neurology, 44*, 330-338.

Gardner-Medwin, D. (1995). New medical classifications and languages. *Developmental Medicine and Child Neurology, 37*, 283-284.

Gerstmann, J. (1940). Syndrome of finger agnosia, disorientation for right and left, agraphia and acalculia. *Archives of Neurology and Psychiatry, 44,* 398-408.

Geschwind, N. (1975). The apraxias; neural mechanisms of disorders of learned movement. *American Scientist, 63*, 188-195.

Gesell, A., & Armatruda, C. (1941). *Developmental Diagnosis Normal and Abnormal Child Development.* New York: Paul B/ Hoeber Inc.

Geuze, R. H. (2003). Static balance and developmental coordination disorder. *Human Movement Science, 22*, 527-548.

Geuze, R. H., & Borger, J. M. A. (1993). Children who are clumsy, five years later. *Adapted Physical Activiy Quarterly, 10*, 10-21.

Geuze, R. H., & Kalverboer A. F. (1994). Tapping a rhythm: a problem of timing for children who are clumsy and dyslexic? *Adapted Physical Activity Quarterly, 11,* 203-213.

Geuze, R. H., & Kalverboer, A. F. (1987). Inconsistency and adaptation in timing of clumsy children. *Journal of Human Movement Studies, 13*, 421-432.

Geuze, R. J., Jongmans, M. J., Schoemaker, M. M., & Smits-Engelsman, B. C. M. (Eds.) (2001) Developmental Coordination Disorder: Diagnosis, Description, Processes and Treatment [Special issue]. *Human Movement Science, 20* (1/2) 7-47.

Ghaziuddin, M., Butler, E., Tsai, L., & Ghaziuddin, N. (1994). Is clumsiness a marker for Asperger syndrome? *Journal of Intellectual Disability Resea*rch, *38*, 519-527.

Gilberg, C. (2003). Deficits in attention, motor control, and perception: a brief review. *Archives of Disease of Childhood, 88*, 904-910.

Gillberg, C., Carlstrom, G., & Rasmussen, P. (1983). Hyperkinetic disorders in seven- year-old children with perceptual, motor and attentional deficits. *Journal of Child Psychology and Psychiatry, 24*, 233-246.

Gillberg, I. C. (1985). Children with preschool minor neurodevelopmental disorders III: Neurodevelopmental profiles at age 20. *Developmental Medicine and Child Neurology, 27*, 3-16.

Gillberg, I. C., & Gillberg, C. (1988). General hyperkinesis: follow –up study from age 7-13 years. *Journal of the American Academy of Child and Adolescent Psychiatry, 27*, 55-59.

Gillberg. I.C., & Gillberg, C. (1989). Children with preschool minor neurodevelopmental disorders. IV Behaviour and school achievement at age 13 *Developmental Medicine and Child Neurology, 31*, 3-13.

Glaser, B. G., & Strauss, A. L. (1967) *The discovery of grounded theory: strategies for qualitative research.* London: Weidenfeld and Nicolson.

Goffman, E. (1963). *Stigma: Notes on the Management of Spoiled Identity.* Englewood Cliffs, NJ: Prentice-Hall

Gonzalez Rothi, L. J., & Heilman, K. M. (Eds.) (1997). *Apraxia: The Neuropsychology of Action.* Hove: Psychology Press.

Goodman, R. (1997). The Strengths and Difficulties Questionnaire: a research note. *Journal of Child Psychology and Psychiatry, 38*, 581-586.

Goodman, R., & Scott, S. (1999). Comparing the Strengths and Difficulties Questionnaire and the Child Behavior Checklist: is small beautiful? *Journal of Abnormal Psychology, 27*, 17-24.

Goodman, R., (2001). Psychometric properties of the Strengths and Difficulties Questionnaire. *Journal of American Academy of Child and Adolescent Psychiatry, 40,* 1337-1345.

Goodman, R., Ford, T., Simmons, H., Gatward, R., & Meltzer, H. (2000). Using the Strengths and Difficulties Questionnaire to screen for child psychiatric disorders in a community sample. *British Journal of Psychiatry, 177,* 534-539.

Gordon, N., & McKinley, I. (1980). *Helping Clumsy Children.* London: Churchill Livingstone.

Grahame, R. (2003). Hypermobility and hypermobility syndrome. in R. Keer, & R. Grahame (Eds.), *Hypermobility Syndrome Recognition and Management for Physiotherapists* (pp. 1-14). Edinburgh: Butterworth Heinmann, Elsvier.

Grahame, R., Bird, H. A., Child, A., Dolan, L., Edwards-Fowler, A., Ferrell, W., Gurley-Green, S., Keer, R., Mansi, E., Murray, K. J., & Smith, E. (2000). The British Society Special Interest Group on Heritable Disorders of Connective Tissue Criteria for the Benign Joint Hypermobility Syndrome. The revised (Brighton 1998) Criteria for the Diagnosis of the BJHS. *Journal of Rheumatology, 27,* 1777-9.

Green, D., Baird, G., Barnett, A. L., Henderson, L., Huber, J., & Henderson, S. E. (2002). The severity and nature of motor impairment in Asperger's syndrome: a comparison with Specific Developmental Disorder of Motor Function. *Journal of Child Psychology and Psychiatry, 43-5,* 655-668.

Grimley, A., & McKinley, I. A. (1977). The Clumsy Child. *Association of Paediatric Chartered Physiotherapists* (APCP).

Gubbay, S. S. (1975). *The clumsy child. A Study of Developmental Apraxic and Agnosic Ataxia.* London: Saunders.

Gubbay, S. S., Ellis, E., Walton, J. N., & Court, S. D. M. (1965). Clumsy children: A study of apraxic and agnosic defects in 21 children *Brain, 88,* 295-312.

Hadders-Algra, M., & Gramsbergen, A. (2003). (Eds.) The Clumsy Child. *Neural Plasticity. Special Issue 10,* (1-2).

Hadders-Algra, M., Huisjes, H. J., & Touwen, B. C. L. (1988). Perinatal risk factors and minor neurological dysfunction: Significance for behaviour and school achievement at nine years. *Developmental Medicine and Child Neurology, 30,* 482-491.

Hagberg, B. (1962). The sequelae of spontaneously arrested infantile hydrocephalus. *Developmental Medicine and Child Neurology, 4,* 583-587.

Hall, D. M. (1988). Clumsy Children. *British Medical Journal, 296,* 375-376.

Hansson, S. L., Svanström Röjvall, A. A., Rastam, M., Gillberg, C., Gillberg, C., & Anckärsater, H. (2005). Psychiatric telephone interview with parents for screening of childhood autism – tics, attention-deficit hyperactivity disorder and other comorbidities (A-TAC). Preliminary reliability and validity. *British Journal of Psychiatry, 187,* 262-267.

Hart, H. (1999). Terminology to benefit children. *Developmental Medicine and Child Neurology, 41*, 651-651.

Harvey, W. J. & Reid, G. (1997). Motor performance of children with Attention-Deficit Hyperactivity Disorder: A preliminary investigation. *Adapted Physical Activity Quarterly, 14*, 189-202.

Harvey, W. J. & Reid, G. (2003). Attention-deficit/hyperactivity disorder: a review of research on movement skill performance and physical fitness. *Adapted Physical Activity Quarterly, 20*, 1-25.

Health Service Journal (1999) June 24th Swallowed whole: beginning of the end for Read Codes. 12-13.

Hellgren, L., Gillberg, C., Gillberg, I. C., & Enerskog, I. (1993). Children with deficits in attention, motor control and perception (DAMP) almost grown up: general health at sixteen years. *Developmental Medicine and Child Neurology, 35*, 881-893.

Hellgren, L., Gillberg, I. C., Bagenholm, A., & Gillberg, C. (1994). Children with deficits in attention, motor control and perception (DAMP) almost grown up: Psychiatric and personality disorders at age 16 years. *Journal of Child Psychology and Psychiatry, 35*, 1255-1271.

Henderson, L., Rose, P., & Henderson, S. (1992). Reaction time and movement time in children with a developmental coordination disorder. *Journal of Psychology, Psychiatry and Allied Disciplines, 33,* 895-905.

Henderson, S. (Ed.) (1994). Special Issue: Developmental Coordination Disorder. *Adapted Physical Activity Quarterly, 1,* 111-114.

Henderson, S. E. (1987). The assessment of "clumsy" children: old and new approaches. *Journal of Child Psychology and Psychiatry, 28*, 511-527.

Henderson, S. E. (1992). Clumsiness or developmental coordination disorder: a neglected handicap. *Current Paediatrics, 2,* 158-162.

Henderson, S. E., & Barnett, A. (1998). Developmental Movement Problems. In J. Rispens, T. van Yperen, & W. Yule (Eds.), *Perspectives on the Classification of Specific Developmental Disorders* (pp. 209-230). Dordrecht: Kluwer Academic Publishers.

Henderson, S. E., & Hall, D. (1982). Concomitants of clumsiness in young school age children. *Developmental Medicine and Child Neurology, 24*, 448-460.

Henderson, S. E., & Henderson, L. (2002). Towards an understanding of developmental coordination disorder in children. The second Rarick lecture. *Adapted Physical Activity Quarterly, 18,* 2-22.

Henderson, S. E., & Sugden, D. A. (1992). *The Movement Assessment Battery for Children.* London: The Psychological Corporation,

Henderson, S. E., Barnett, A. L., & Henderson, L. (1994). Visuospatial difficulties and clumsiness: on the interpretation of conjoined deficits. *Journal of Child Psychology and Psychiatry, 35,* 961-969.

Henderson, S. E., May, D. S., & Umney, M. (1989). An exploratory study of goal-setting behaviour, self-concept and locus of control in children with movement difficulties. *European Journal of Special Needs Education, 4,* 1-15.

Hill, E. L. (1998). A dyspraxic deficit in specific Language Impairment and Developmental Coordination Disorder? Evidence from hand and arm movements. *Developmental Medicine and Child Neurology, 40,* 388-395.

Hill, E. L. (2001). Non-specific nature of specific language impairment: a review of the literature with regard to concomitant motor impairments. *International Journal of Language & Communication Disorders, 36,* 149-171.

Hill, E. L., Bishop, D. V. M., & Nimmo-Smith, I. (1998). Representational gestures in Developmental Coordination Disorder and specific language impairment: Error-types and the reliability ratings. *Human Movement Science, 17,* 655-678.

Hiscock, M., & Chapieski, L. (2004). Hand preference, manual asymmetry and manual skill. in D. Dewey & D. E. Tupper (Eds.), *Developmental Motor Disorders: A Neuropsychological Perspective* (pp. 353-388). New York: Guilford Press.

Hoare, D. (1994). Subtypes of Developmental Coordination Disorder. *Adapted Physical Activity Quarterly, 11,* 158-169.

Horak, F. B., Shumway-Cook, A., Crowe, T. K., & Black, F. O. (1988). Vestibular function and motor proficiency in children with impaired hearing or with a learning disability and motor impairments. *Developmental Medicine and Child Neurology, 30,* 64-79.

Hulme, C., Smart, A., Moran, G., & McKinlay, I. (1984). Visual, kinaesthetic and cross-modal judgements of length by clumsy children: a comparison with young normal children. *Child: Care, Health and Development, 20,* 117-125.

Huson, S. M., Harper, P. S., & Compston, D. A. S. (1988). Von Recklinghausen neurofibromatosis: a clinical and population study in south east Wales. *Brain, 111,* 1355-1381.

Hyman, R. (1989). *The Pan Dictionary of Famous Quotations.* London: Pan Books.

Hyman, S. L., Shores, A., & North, K. N. (2005). The nature and frequency of cognitive deficits in children with neurofibromatosis type 1. *Neurology, 65,* 1037-1044.

Illingworth, R. S. (1963). The clumsy child. In M. Bax & R. MacKeith (Eds.), *Minimal Cerebral Dysfunction. Clinics in Developmental Medicine,* No 10. London: Spastics International Medical Publications with Heinemann Medical Books Ltd.

Iloeje, S. O. (1987). Developmental apraxia among Nigerian children in Enugu, Nigeria. *Developmental Medicine and Child Neurology, 29*, 502-507.

Ingram, T. T. S. (1963). Chronic brain syndromes in childhood other than cerebral palsy, epilepsy and mental defect. In M. Bax & R. MacKeith (Eds.), *Minimal Cerebral Dysfunction. Clinics in Developmental Medicine,* No 10. London: Spastics International Medical Publications with Heinemann Medical Books Ltd.

Ingram, T. T. S. (1984). A historical review of the definition and classification of the cerebral palsies. In F. Stanley & E. Alberman (Eds.), *The Epidemiology of the Cerebral Palsies* (pp 1-11) [Monograph] *Clinics in Developmental Medicine, 87.* Oxford: Spastics International Medical Publications with Blackwell Scientific.

Jansson, A., Saartok, T., Werner, S., & Renström, P. (2004). General joint laxity in 1845 Swedish school children of different ages: age- and gender-specific distributions. *Acta Paediatrica, 93*, 1202-1206.

Johnson, M. H. (1997). *Developmental Cognitive Neuroscience*. Oxford: Blackwell.

Jongmans, M. J. (1993). *Perceptuo-motor competence in prematurely born children at school age: Neurological and psychological aspects*. Doctoral thesis. University of London.

Jongmans, M., Henderson, S. E., de Vries, L., & Dubowitz, L. (1993). Duration of periventricular densities in preterm infants and neurological outcome at 6 years of age. *Archives of Disease in Childhood, 69*, 9-13.

Jongmans, M., Mercuri, E., Dubowitz, L., & Henderson, S. (1998). Perceptual-motor difficulties and their concomitants in six-year-old children born prematurely. *Human Movement Science, 17,* 629-653.

Kadesjo, B., & Gillberg, C. (1998). Attention deficits and clumsiness in Swedish 7-year-old children. *Developmental Medicine and Child Neurology, 40*, 796-804.

Kadesjo, B., & Gillberg, C. (2001). The co-morbidity of ADHD in the general population of Swedish school-children. *Journal of Child Psychology and Psychiatry, 42*, 487-492.

Kalverboer, A. F., van Praag, H. M., & Mendlewica, J. (1978). *Minimal Brain Dysfunction: Fact or Fiction.* Basel, Switzerland: S. Karger.

Kaplan, B. J. (2005). *Comorbidity, co-occurrence, continuum: What's in a name?* Paper presented at 6th International Conference on Developmental Coordination Disorder Trieste, Italy.

Kaplan, B. J., Dewey, D., Crawford, S., & Wilson, B. N. (2001). The term *comorbid*ity is of questionable value in reference to developmental disorders: data and theory. *Journal of Learning Disabilities, 34,* 555-565.

Kaplan, B. J., Wilson, B. N., Dewey, D., & Crawford, S. G. (1998). DCD may not be a discrete disorder. *Human Movement Science, 17*, 471-490.

Kaplan, B., Myers, G. R., & Koogler, T. K. (2004). Parents'/guardians' views on donation after cardiac death. *Critical Care Medicine, 32*(12). Supplement A89.

Kavey, R. W., Daniels, S. R., Lauer, R. M., Atkins, D. L., Hayman, L. L., & Taubert, K. (2003). American heart association guidelines for primary prevention of atherosclerotic cardiovascular disease beginning in childhood. *The Journal of Pediatrics, 142*, 368-372.

Kelso, J. A. S. (1982). Concepts and Issues in Human Motor Behavior: Coming to Grips with the Jargon. In J. A. S. Kelso (Ed.), *Human Motor Behavior: An Introduction.* New Jersey: Lawrence Erlbaum Associates Inc.

Kelso, J. A. S., & Tuller, B. (1981). Toward a theory of apractic syndromes. *Brain and Language, 12,* 224 - 245.

Kendall, P. C., & Clarkin, J. F. (1992). Introduction to special Section; Comorbidity and treatment implications. *Journal of Clinical and Consulting Psychology, 60*, 833-834.

Keogh, J. F. (1968). Incidence and severity of awkwardness among regular school boys and educationally subnormal boys. *Research Quarterly, 39*, 806-808.

Kinsbourne, M., & Warrington, E. K. (1963). The developmental Gerstmann syndrome. *Archives of Neurology, 8,* 490-501.

Kirby, A., Davies, R., & Bryant, A. (2005). Hypermobility Syndrome and developmental co-ordination disorder. *International Journal of Therapy and Rehabilitation, 12*(10) 431-437.

Klin, A., Volkmar, F. R., Sparrow, S. S., Cicchetti, D. V., & Rourke, B. P. (1995). Validity and neuropsychological charactreization of Asperger Syndrome: Convergence with Non Verbal Learning Disabilities Syndrome. *Journal of Child Psychology and Psychiatry, 36*, 1127-1140

Knobloch, H., & Pasamanick, B. (1959). Syndrome of minimal damage in infancy. *Journal of the American Medical Association, 170*, 1384-1387.

Kooistra, L., Stemerdink, N., van der Meere, J., Vulsma, T., & Kalverboer, A. F. (2001). Behvarioual correlates of early-treated congenital hypothyroidism. *Acta Paediatrica, 90*, 1141-6.

Kramer, F., & Pollnow, H. (1932). Uber eine hyperkinetische Erkrankung im Kindesalter. *Monatsschrift fur Psychiatrie und Neurologie, 82*, 1-40.

Landgren, M., Peteresson, R., Kjellman, B., & Gillberg, C. (1996). ADHD, DAMP and other neurodevelopmental/psychiatric disorders in six-year-old children: Epidemiology and co-morbidity. *Developmental Medicine and Child Neurology, 38*, 891-906.

Laszlo, J. I., & Bairstow, P. J. (1983). Kinaesthesis: Its measurement, training and relationship to motor control. *Quarterly Journal of Experimental Psychology, 35a,* 411-421.

Leeds Consensus Statement (2006). Economic Science Research Council Seminar Series: Developmental Coordination Disorder as a Specific Learning Difficulty; 2004-2006. Principal Investigator: D. A. Sugden.

Levene, M., Dowling, S., Graham, M., Fogelman, K., Galton, M., & Phillips, M. (1992). Impaired motor function (clumsiness) in 5 year old children: correlation with neonatal ultrasound scans. *Archives of Disease of Childhood, 67,* 687-690.

Lipson, A., Edwards, P., & Logan, G. S. (2003). Occupational therapy and physiotherapy for developmental coordination disorder. (Protocol) *The Cochrane Database of Systematic Reviews*, Issue I. Art. No.: CD004256. DOI:10.1002/14651858.CD004256.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 24,* 659-685.

Lord, C., Rutter, M., Goode, S., Heemsbergen, J., Jordan, H., Mawhood, L. & Schopler, E. (1989). Autism Diagnostic Observation Schedule: a standardised observation of communicative and social behaviour. *Journal of Autism and Developmental Disorders, 19,* 185-212.

Lord, R. & Hulme, C. (1988). Visual perception and drawing ability in normal and clumsy children. *British Journal of Developmental Psychology, 6,* 1-9.

Losse, A., Henderson, S. E., Elliman, D., Hall, D., Knight, E., & Jongmans, M. (1991). Clumsiness in Children - do they grow out of it? A 10-year follow-up study. *Developmental Medicine and child Neurology, 33,* 55-68.

MacCurdy, E. (1938). Translation Manuscript H (2179), Library of the Institut de France in: The Notebooks of Leonardo da Vinci, Vol. 1 Chapter 1 London

Mackeith, R. C. (1968). Maximal clarity on neurodevelopmental disorders. *Developmental Medicine and Child Neurology, 10,* 143-144.

Macnab, J. J., Miller, L. T., & Polatajko, H. J. (2001). The search for subtypes of DCD: is cluster analysis the answer? *Human Movement Science, 20,* 49-72.

Maillard, S. & Murray, K. J. (2003). Hypermobility Syndrome in Children. In R. Keer & R. Grahame (Eds.), *Hypermobility Syndrome* (pp 33- 50). Edinburgh: Butterworth, Heinmann.

Manly, B. F. J. (1994). Cluster Analysis. In B. F. J. Manly (Ed.) *Multivariate Statistical Methods A Primer* (pp. 128-145). London: Chapman & Hall.

Marchiori, G. E., Wall, A. E., & Bedingfield, E. W. (1987). Kinematic analysis of skill acquisition in physically awkward boys. *Adapted Physical Activity Quarterly, 4*, 305-315.

Mayes, S. D., Calhoun, S. L., & Crites, D. L. (2001). Does DSM-IV Aspergers's disorder exist? *Journal of Abnormal Child Psychology, 29*, 263-271.

McCarron, L. T. (1982). MAND – *McCarron Assessment of Neuromuscular Development (revised ed.)*. Dallas, TX: Common Market Press.

McKinley, I. (1996). The education act 1993: working with health services to implement the Code of Practice. *Child: Care, Health and Development, 22*, 19-30.

Medical Research Council (1943). *Aids to the Investigation of Peripheral Nerve Injuries. War memorandum No 7*, 2nd Ed. London: HMSO.

Miller, N. (1988). The assessment of limb dyspraxia. *Clinical Rehabilitation, 2,* 177-181.

Missiuna, C. (2001). (Ed.) *Children with Developmental Coordination Disorder: Strategies for Success.* New York: The Howarth Press. Co-published as a special issue of *Physical and Occupational Therapy in Pediatrics, 20,* (2/3).

Missiuna, C., & Polatajko, H. (1995). Developmental dyspraxia by any other name: are they all just clumsy children? *American Journal of Occupational Therapy, 49*, 619-627.

Miyahara, M. (1994). Subtypes of students with learning disabilities based upon gross motor functions. *Adapted Physical Activity Quarterly, 11*, 368-382.

Miyahara, M., & Register, C. (2000). Perceptions of three terms to describe physical awkwardness in children. *Research in Developmental Disabilities, 21*, 367-376.

Murray, K. J. (2006). Hypermobility disorders in children and adolescents. Best Practice and Research. *Clinical Rheumatology, 20*, 329-351

Murray, K. J., & Woo, P. (2001). Benign joint hypermobility in childhood. (Editorial). *Rheumatology, 40*, 489-491.

National Health Service Executive (1999). *Clinical Governance, Quality in the New NHS.* Bristol, UK, Author.

Naville, S. (1970). Psycho-motor therapy. *Remedial Education, 9*, 13-13.

Nichols, P., & Chen, T. (1980). *Minimal Brain Dysfunction: A Prospective Study.* Hillsdale, N.J: Lawrence Erlbaum Associates.

Nicolson, R. I., & Fawcett, A. J. (1990). Automaticity: A new framework for dyslexia research. *Cognition, 35,* 159-182.

Nicolson, R. I., Fawcett, A. J., & Dean, P. (1995). Time estimation of deficits in developmental dyslexia: evidence of cerebellar involvement. *Proceedings of Royal Society of London (Series B) 259*, 43-47.

Normative Data from Britain (n.d.) Accessed 24th April 2006 from www.sdqinfo.com/bbl.html

Norussis, M. J. (2002). *SPSS for Windows: Base System User's Guide*: *Release 11.0.* Chicago: SPSS Inc.

Norussis, M. J. (2005) *SPSS for Windows: Version 12.0 for Windows.* Chicago: SPSS Inc.

Nutton, V. (personal communication, 2005). Welcome Trust Centre History of Medicine. University College London.

O'Brian, B., Cermak, S. A, & Murray, E. (1988). The relationship between visual-perceptual motor abilities and clumsiness in children with and without learning disabilities. *American Journal of Occupational Therapy, 42*, 359-363.

O'Hare, A., & Khalid, S. (2002). The association of abnormal cerebellar function in children with developmental coordination disorder and reading difficulties. *Dyslexia, 8,* 234-248.

O'Hare, A. E., & Brown, J. K. (1989). Childhood dysgraphia: Part 2. A study of hand function. *Child: Care, Health and Development, 15*, 151-166.

Oppenheim, A. N. (1992). *Questionnaire Design Interviewing and Attitude Measurement* (2nd ed.). New York, NY: Pinter Publishers.

Orton, S. T. (1937). *Reading, Writing and Speech Problems in Children.* New York: Norton.

Oxbury, J. M., & Oxbury, S. M. (1996). Organization and Features of Dysfunction. In D. J. Weatherall, J. G. G. Ledingham & D. A. Warrell (Eds.), *Oxford Textbook of Medicine* (3rd ed.) (pp. 3845-3856). Oxford: Oxford University Press.

Paine, R. S. (1962). Minimal chronic brain syndromes in children. *Developmental Medicine and Child Neurology, 4,* 21-27.

Paine, R. S., Werry, J. S., & Quay, H. C. (1968). A study of "minimal cerebral dysfunction" *Developmental Medicine and Child Neurology, 10*, 505-520.

PeBenito, R. (1987). Developmental Gertsmann Syndrome: Case report and review of the literature. *Developmental and Behavioural Pediatrics, 8,* 229-232.

Pellecchia, G. L. (2003). Postural sway increases with attentional demands of concurrent cognitive task. *Gait and Posture, 18*, 29-34.

Pellegrino, L. (1995). Cerebral palsy: a paradigm for the developmental disabilities. Annotation. *Developmental Medicine and Child Neurology, 37*, 834-839.

Peters, J. M., & Wright, A. M. (1999). Development and evaluation of a group physical activity programme for children with developmental coordination disorder: An interdisciplinary approach. *Physiotherapy Theory and Practice, 15*, 203-216.

Peters, J. M., Barnett, A. L., & Henderson, S. E. (2001). Clumsiness Dyspraxia and Developmental Coordination Disorder: How do health and educational professionals in the UK define the terms? *Child: Care Health and Development 27*, 399-412.

Peters, J. M., Henderson, S. E., & Dookun, D. (2004). Provision for children with developmental coordination disorder (DCD): Audit of the service provider. *Child: Care Health and Development, 30*(5), 463-479.

Pette, D., & Vrbova, G. (1995). Neural control of phenotype expression in mammalian muscle fibres. *Muscle Nerve, 8*, 676 - 689.

Phelps, W. M. (1949). Description and differentiation of types of cerebral palsy. *Nervous Child, 8,* 107-127.

Piek, J. P., Pitcher, T. M., & Hay, D. A. (1999). Motor coordination and kinaesthesis in boys with attention deficit-hyperactivity disorder. *Developmental Medicine and Child Neurology, 41*, 159-165.

Pitcher, T. M., Piek, J.P., & Hay, D. A. (2003). Fine and gross motor ability in males with ADHD *Developmental Medicine and Child Neurology, 45*, 525-535.

Pless, M., & Carlsson, M. (2000). Effects of motor skills intervention on Developmental Coordination Disorder: A meta-analysis. *Adapted Physical Activity Quarterly, 17*, 381–401.

Polani, P. E. (1960). Is paediatric neurology a good idea? Child Neurology and Cerebral Palsy: A Report of an International Study Group. *Little Club Clinics in Developmental Medicine, 2,* 5-8. Medical Advisory Committee of the National Spastics Society.

Polatajko, H. J. (1999). Developmental Coordination Disorder (DCD): Alias the clumsy child syndrome. In K. Whitmore, H. Hart, & G. Williams (Eds.), *A Neurodevelopmental Approach to Specific Learning Disorders* (pp. 119-133) London: Mac Keith Press.

Polatajko, H. J., Fox, M., &Missiuna, C. (1995) An international consensus on children with Developmental Coordination Disorder. *Canadian Journal of Occupational Therapy, 62,* 3-6.

Polatajko, H. J., Mandich, A. D., Miller, L. T., Macnab, J. J. (2001). Cognitive orientation to daily occupational performance (CO-OP). Part 2: The evidence. *Physical and Occupational Therapy in Pediatrics, 20,* 83-106.

Portwood, M. (1996). *Developmental dyspraxia: a practical manual for parents and professionals.* Durham: Durham County Council.

Portwood, M. (1999). *Developmental dyspraxia: Identification and Intervention.* London: David Fulton.

Posserud, M. B., Lundervold, A. J., & Gillberg, C. (2006). Autistic features in a total population of 7-9-year-old children assessed by the ASSQ (Autism Spectrum Screening Questionnaire). *Journal of Child Psychology and Psychiatry, 47,* 167-175.

Powell, R. P., & Bishop, D. V. M. (1992). Clumsiness and perceptual problems in children with specific language impairment. *Developmental Medicine and Child Neurology, 34,* 755-765.

Prechtl, H. F. R., & Stemmer, C. H. (1962). The choreiform syndrome in children *Developmental Medicine and Child. Neurology, 4,* 119-127.

Purvis, R. J. (1989). The clumsy child. *British Journal of Orthoptics, 46,* 98-101.

Ramus, F., Pidgeon, E., & Frith, U. (2003). The relationship between motor control and phonology in dyslexic children. *Journal of Child Psychology and Psychiatry, 44,* 712-722.

Random number generator (n.d.) Accessed June 10th 2003 from www.random.org

Rasmussen, P., & Gillberg, C. (2000). Natural outcome of ADHD with developmental coordination disorder at age 22 years: a controlled, longitudinal, community-based study. *Journal of the American Academy of Child and Adolescent Psychiatry, 39,* 1424-1431.

Reuben, R. N., & Bakwin, H. (1968). Developmental clumsiness. *Paediatric Clinics of North America 15,* 601-610.

Riach, C. L., & Starkes, J. L. (1993). Stability limits of quiet standing postural control in children and adults. *Gait and Posture, 1,* 105-111.

Ripley, K., Daines, B., & Barrett, J. (1997). *Dyspraxia. A Guide for Teachers and Parents.* London: David Fulton.

Rispens, J., van Yperen, T. A., & Yule, W. (Eds.) (1998). *Perspectives on the Classification of Specific Developmental Disorders.* Dordrecht: Kluwer Academic Publishers.

Rösblad, B., & von Hofsten, C. (1994). Repetitive goal-directed arm movements in children with developmental coordination disorders: role of visual information. *Adapted Physical Activity Quarterly, 11,* 190-202.

Rosenbaum, P. (2006). Commentary: Infantile posture asymmetry and osteopathic treatment: a randomised therapeutic trial. *Developmental Medicine and Child Neurology, 48,* 4-4.

Roy, E. A. (1978). Apraxia: a new look at an old syndrome. *Journal of Human Movement Studies, 4,* 191-210.

Russek, L. N. (1999). Hypermobility syndrome. *Physical Therapy, 79*, 591-599.

Rutter, M. (1977). Brain damage syndromes in childhood: concepts and findings. *Journal of Psychiatry, 18*, 1-21.

Rutter, M. (1981). Psychological sequelae of brain damage in children. *American Journal of Psychiatry, 138*, 1533-1544.

Rutter, M. (1982). Syndromes attributed to "minimal brain dysfunction" in childhood. *American Journal of Psychiatry, 139*, 21-33.

Rutter, M. (1998). Preface In: J. Rispens, T. van Yperen,. W. Yule (Eds.), *Perspectives on the Classification of Specific Developmental Disorders*(pp ix-xviii). Dordrecht: Kluwer Academic Publishers.

Schachar, R. J. (1986). Hyperkinetic syndrome: historical development of the concept. In E. A. Taylor (Ed.), *The overactive Child.* [Monograph] *Clinics in Developmental Medicine, 97,* 19-40. Oxford: Spastics International Medical Publications.

Schaffer, D. (1978). "Soft" neurological signs and later psychiatric disorder: A review. *Journal of Child Psychology and Psychiatry, 19*, 63-65.

Schoemaker, M. M., & Kalverboer, A. F. (1994). Social and affective problems of children who are clumsy: How early do they begin? *Adapted Physical Activity Quarterly, 11*, 130-140.

Schoemaker, M. M., Hijlkema, M. G. J., & Kalverboer, A. F. (1994). Physiotherapy for clumsy children: an evaluation study. *Developmental Medicine and Child Neurology, 36*, 143-155.

Serrien, D. J., Nirkko, A. C., Loher, T. J., Lovblad, K. O., Burgunder, J. M., & Wiesendanger, M. (2002). Movement control of manipulative tasks in patients with Gilles de la Tourette syndrome. *Brain, 125*, 290-300.

Shumway-Cook, A., & Wollacott, M. (1985). The growth of stability: postural control from a developmental perspective. *Motor Behavior, 17,* 131-147.

Sims, K., Henderson, S. E., Morton, J., & Hulme, C. (1996). The remediation of clumsiness 11: is kinaesthesis the answer? *Developmental Medicine and Child Neurology, 38*, 988-887.

Singer, C. (1928). *A short History of Medicine*. Oxford: Clarendon Press.

Skuse, D., et al. (2004). The Developmental, Dimensional and Diagnostic Interview (3Di): a novel computerized assessment for autism spectrum disorders. *Journal of the Ameriacan Academy of Child and Adolescent Psychiatry, 43*, 548-558.

Smith, I. (2005). Motor problems in children with autistic spectrum disorders. In D. Dewey & D. E. Tupper (Eds.), *Developmental Motor Disorders: A Neuropsychological Perspective* (pp 152-168). New York:Guilford Press.

Square, P. A., Roy, E. A., & Martin, R. E. (1997). Apraxia of speech: another form of disturbance. In: L. J. Gonzalez Rothi & K. M. Heilman (Eds.), *Apraxia: The Neuropsychology of Action.* East Sussex: Psychology Press.

Stanovich, K. E. (1994). Annotation: Does dyslexia exist? *Journal of Child Psychology and Psychiatry, 35,* 579-595.

Stephenson, E., Mckay, C., & Chesson, R. (1990). An investigative study of early developmental factors in children with motor/learning difficulties. *British Journal of Occupational Therapy, 53,* 4-6.

Stephenson, E., McKay, C., & Chesson, R. (1991). The identification and treatment of motor/learning difficulties: parents' perceptions and the role of the therapist. *Child; Care, Health and Development, 17,* 91-113.

Still, G. F. (1902). The Coulstonian Lectures on some abnormal psychical conditions in children. *Lancet, 1,* 1008-1012; 1077-1082; 1163-1168.

Stott, D. H., Moyes, F. A, & Henderson, S. E. (1984). *Test of Motor Impairment.* Henderson Revision. Guelph, Ontario: Brook Educational.

Strauss, A. A., & Lehtinen, L. E. (1947). *Psychopathology and Education of the Brain-Injured Child.* New York: Grune and Stratton.

Strauss, A., & Kepart, N. C. (1955). *Psychopathology and Education of the Brain-Injured Child Vol II, Progress and Theory Clinic.* New York: Grune and Stratton.

Sugden, D. A., & Chambers, M. E. (1998). Intervention approaches and children with developmental coordination disorder. *Paediatric Rehabilitation, 2,* 139-147.

Sugden, D. A., & Chambers, M. E. (2005). (Eds.) *Children with Developmental Coordination Disorder.* London: Whurr Publishers.

Sugden, D. A., & Wright, H. C. (1998). Motor Coordination Disorders in Children. *Developmental Clinical Psychology and Psychiatry,* Vol. 39. London: Sage Publications.

Taylor, D. C., & McKinlay, I. A. (1979). What type of thing is being clumsy?. *Child: Care Health & Development, 5,* 167-175.

The Compact Oxford English Dictionary (1991). (2nd ed.) Oxford: Clarendon Press.

The Oxford Dictionary of New Words (1997). Oxford: University Press.

Thelen, E., Kelso, J. A., & Fogel, A. (1987.) Self-organising systems and infant motor development. *Developmental Review, 7,* 39-65.

Touwen, B. C. L. (1979). *Examination of the Child with Minor Neurological Dysfunction* (2nd ed), [Monograph] *Clinics in Developmental Medicine, 71.* Oxford: Spastics International Medical Publications with Heinemann Medical.

Touwen, B. C. L., & Prechtl, H. (1970). *The Neurological Examination of the Child with Minor Nervous Dysfunction.* [Monograph] *Clinics in Developmental Medicine, 38.* London: S.I.M.P. with Heinemann Medical.

Tredgold, A. F. (1908). *Mental Deficiency (Amentia).* New York: W. Wood.

Tryon, R. C. (1939). *Cluster Analysis: Correlation Profile and Orthometric Analysis for the Isolation of Unities in Mind and Personality.* Ann Arbor, MI: Edwards Brother.

Ulrich, D. A. (2000). *Test of Gross Motor Development* (2nd ed.) *Examiner's manual.* Austin, TX: Pro-ed.

Unwin, J. (1995). Current perspectives on minimal neurological dysfunction. *Australian Journal of Physiotherapy, 41,* 9-12.

Van der Giesson, L. J., Liekens, D., Rutgers, K. J., Hartman, A., Mulder, P. G., & Oranje, A. P. (2001). Validation of Beighton score and prevalence of connective tissue signs in 773 Dutch children. *Journal of Rheumatology, 28,* 2726-2730.

Visser, J. (2003). Developmental coordination disorder: a review of research on subtypes and comorbidities. *Human Movement Science, 22,* 479-493.

Visser, J., Geuze, J. H., & Kalverboer, A. F. (1998). The relationship between physical growth, the level of activity and the development of motor skills in adolescence: Difference between children with DCD and controls. *Human Movement Science, 17,* 573-605

Volman, M. C. J. M., & Geuze, R. H. (1998). Relative phase stability of bimanual and visuomanual rhythmic coordination patterns in children with a Developmental Coordination Disorder. *Human Movement Science, 17,* 541-572.

Wall, A. E., Reid, G., & Paton, J. (1990). The syndrome of physical awkwardness. In G. Reid (Ed). *Problems in Movement Con*trol. Amsterdam: Elsevier.

Wallen, M., Bonney, M. A., & Lennox, I. (1996). *The Handwriting Speed Test.* Adelaide: Helios.

Walton, J. N. (1961). Clumsy Children. *Spastics Quarterly 10,* 9-9.

Walton, J. N. (1963). Clumsy children. In M. Bax & R. MacKeith (Eds.), *Minimal Cerebral Dysfunction.* [Monograph] *Clinics in Developmental Medicine, 10.* London: Spastics International Medical Publications with Heinemann Medical Books Ltd.

Walton, J. N., Ellis, E., & Court, S. D. M. (1962). Clumsy Children: a study of developmental apraxia and agnosia. *Brain, 85*, 603-612.

Wann, J. P., Mon-Williams, M., & Rushton, K. (1998). Postural control and co-ordination disorders: the swinging room revisited. *Human Movement Science, 17*, 491-513.

Warnock, H. M. (1978). *Special Educational Needs: Report of the Committee of Enquiry into the Education of Handicapped Children and Young People.* London: HMSO.

Watter, P., & Bullock, M. I. (1987). Patterns of improvement in neurological functioning in children with minimal cerebral dysfunction with physiotherapy intervention. *Australian Journal of Physiotherapy, 33*, 215 – 224.

Wechsler, D. (1992). *Wechsler Intelligence Scale for Children (WISC)* (3rd ed.). London: Psychological Corporation.

Wedell, K. (1972). *Perceptuo-Motor Disabilities in Children.* London: John Wiley and Sons.

Whitmore, K., & Bax, M. (1999). What do we mean by SLD? A Historical Perspective. in K. Whitmore, H. Hart & G. Willems (Eds.), *A Neurodevelopmental Approach to Specific Learning Difficulties* (pp 1- 23). [Monograph] *Clinics in Developmental Medicine,* No. 143. London: Mac Keith Press distributed by Cambridge University Press.

Williams, H. G., & Ho, L. (2004). Balance and postural control across the lifespan. in D. Dewey & D. E. Tupper (Eds.), *Developmental Motor Disorders: A Neuropsychological Perspective* (pp. 211-234). New York: Guilford Press.

Williams, H. G., Fisher, J. M., & Tritschler, K. A. (1983). Descriptive analysis of static postural control in 4- 6- and 8-year old normal and motorically awkward children. *American Journal of Physical Medicine, 62*,12-26.

Wilson, B., Dewey, D., & Campbell, A. (1998). *The Developmental Coordination. Disorder Questionnaire.* Calgary: Author.

Winter, D. A. (1995). Human balance and posture control during standing and walking. *Gait & Posture, 3,* 193-214.

Wolfe, P. H., & Hurwitz, J. (1966). The choreiform syndrome. *Developmental Medicine and Child Neurology, 8,* 160-165.

Working Party (1999). *Dyslexia, Literacy and Psychological Assessment.* Report of a Working Party of the Division of Educational and Child Psychology 1-125. Leicester, UK: The British Psychological Society.

World Health Organization (1948). *International Statistical Classification of Diseases and Related Health Problems (ICD-6).* Geneva: WHO.

World Health Organization (1980). *The International Classification of Impairments, Disabilities and Handicaps.* Geneva: WHO.

World Health Organization (1992). *The Classification of Mental and Behavioural Disorders: Clinical Descriptions and Diagnostic Guidelines (ICD-10).* Geneva: WHO.

World Health Organization (2001). *International Classification of Functioning, Disability and Health (ICF)* World Health Assembly. 54/6 Press Release 22nd May: Geneva: WHO.

World Health Organization (2002) *International Classification of Functioning, Disability and Health (ICF).* Geneva: World Health Organization.

Wright, H. C., & Sugden, D. A (1996b). A two step procedure for the identification of children with developmental coordination disorder in Singapore. *Developmental Medicine and Child Neurology, 38*(12), 1099-1106.

Wright, H. C., & Sugden, D. A. (1996a). The nature of developmental coordination disorder: Inter- and intragroup difference. *Adapted Physical Activity Quarterly, 13*, 357-371.

Zoia, S., Barnett, A., Wilson, P., & Hill, E. L. (2006). Developmental coordination disorder: Current issues. *Child: Care, Health and Development.* In press.

Appendices

Appendix 1. *Ethics Committee Approval*

Institute of Child Health
and Great Ormond Street Hospital for Children NHS Trust
UNIVERSITY COLLEGE LONDON

30 Guilford Street, London, WC1N 1EH Telephone 020 7242 9789 Fax: 020 7813 8234

The child first and always

27 March 2003

Mrs JM Peters
Clinical Specialist Physiotherapist
Physiotherapy
GOS

Dear Ms Peters,

Title: Developmental Coordination Disorder (DCD): One syndrome with many subtypes, or one 'symptom' of may different syndromes?
R&D registration number: 03NS02
Protocol number/version: N/A

Notification of ethical approval

The above research has been given ethical approval after review by the Great Ormond Street Hospital for Children NHS Trust / Institute of Child Health Research Ethics Committee subject to the following conditions.

1. Your research must commence within twelve months of the date of this letter and ethical approval is given for a period of 24 months from the commencement of the project. If you wish to start the research more than twelve months from the date of this letter or extend the duration of your approval you should seek Chairman's approval.
2. You must seek Chairman's approval for proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature, eg. using the same procedure(s) or medicinal product(s). Each research project is reviewed separately and if there are significant changes to the research protocol, for example in response to a grant giving body's requirements you should seek confirmation of continued ethical approval.
3. Researchers are reminded that REC approval does not imply approval by the GOS Trust. Researchers should confirm with the R&D office that all necessary permissions have been obtained before proceeding.
4. It is your responsibility to notify the Committee immediately of any information which would raise questions about the safety and continued conduct of the research.
5. On completion of the research, you must submit a report of your findings to the Research Ethics Committee. You may also be required to submit annual reports.
6. Specific conditions pertaining to the approval of this project are:
   - The use of the enclosed standard consent forms for the research. A copy of the signed consent form must be placed in the patient's clinical records and a copy must be kept by you with the research records.

Yours sincerely

Orlagh Sheils
Administrator to the Research Ethics Committee

Appendix 2. *Local Research Ethics Committee Conversion to Multi-centre Project with No Local Investigator*

Great Ormond Street Hospital for Children NHS Trust / The Institute of Child Health Local Research Ethics Committee
Institute of Child Health
30 Guilford Street
London
WC1N 1EH

Tel 020 7905 2620
Fax 020 7905 2201
Email l.howe@ich.ucl.ac.uk

22nd March 2004

Judith Peters
Clinical Specialist Physiotherapist
Level 5, Frontage Building
GOSH

Dear Judith,

Full title of study:
Developmental Coordination Disorder (DCD): One syndrome with many subtypes, or one 'symptom' of many different syndromes
REC reference number:
03NS02

Amendment:
Extension of recruitment to the Royal Free and Ealing Community Hospital; conversion of the study to a multi-centre project with no local investigator

The above amendment was reviewed by a Sub-Committee of the Great Ormond Street Hospital for Children NHS Trust/Institute of Child Health Research Ethics Committee at the meeting held on 18th March 2004.

Ethical opinion

The members of the Committee present gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Site-specific issues

As this is a study with no local investigators, there is no need to inform Local Research Ethics Committees of this amendment.

Approval of host organisations

Local principal investigators or research collaborators should notify their host organisations of this amendment and check whether it affects local management approval of the research.

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are:
Dr V Larcher (Consultant Paediatrician, Chair of GOSH/ICH REC)
Dr E Main (Physiotherapy Research Coordinator)

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

Yours sincerely,

Laura Howe
Research Ethics Coordinator

Copy to: R&D Office, ICH

Appendix 3. *Information Sheet*

We would like to include your child in a study about children's movement.

Aim of the study.

Most children easily learn to move about and use their hands and fingers for everyday activities. However there are children who appear otherwise healthy, and bright but have great difficulty learning to tie shoelaces, do up buttons or write. Others have more difficulty with hopping, jumping, riding a bicycle or catching a ball. All of these difficulties, but especially handwriting, may make it hard to get on in school. We want to learn more about the different kinds of movement difficulties but also about children who find movement no problem.

Why is the study being done?

The study is being done to learn more about children's movement so that we may be able to develop the best treatment programmes to help those children with movement difficulties.

How is the study being done?

If you are happy for your child to take part, your child will be invited to attend for a physiotherapy assessment at Great Ormond Street Hospital for Children. The assessment should involve a morning or afternoon session with a short break. Your child will be asked do some every day activities, such as jumping, balancing like he/she does when playing with friends. Your child will also do some simple pencil and paper games. Your child is likely to find the activities fun and to enjoy the session.

Are there any risks and discomforts?

No risk or discomfort to your child is foreseen.

What are the potential benefits?

The results we obtain will help us to plan treatments in the best possible way for those children who have difficulty with everyday actions.

Who will have access to the research records?

Only the researchers and a representative of the research ethics committee will have access to the data collected during this study. The use of some types of personal information is safe guarded by the Data Protection Act 1998 (DPA). The DPA places an obligation on those who record or use personal information, but also gives rights to people about whom information is held. If you have any questions about data protection, contact the Data Protection officer via the switchboard on 020 7405 9200 extension 5217

What are the arrangements for compensation, should any harm come to the subject?

This project has been approved by an independent research ethics committee who believe that it is of minimal risk to you. However, research can carry unforeseen risks and we want you to be informed of your rights in the unlikely event that any harm should occur as a result of taking part in this study.

No special compensation arrangements have been made for this project but you have the right to claim damages in a court of law. This would require you to prove fault on the part of the Hospital and/or manufacturer involved.

Do I have to take part in this study?

If you decide now or at a later stage that you do not wish to participate in this research project, that is entirely your right and will not in any way change the way your child is treated.

Researcher who will have contact with the family

Judith Peters, Clinical Specialist Physiotherapist
Physiotherapy Department, Great Ormond Street Hospital for Children.
Telephone: 020 7829 8610

Who do I speak to if problems arise?

If you have any complaints about the way in which this research project has been or is being conducted, please in the first instance discuss them with the researcher. If the problems are not resolved, or you wish to comment in any way, please contact the chairman of the Research Ethics Committee, by post via the Research and Development Office, Institute of Child Health, 30 Guildford Street, London, WC1N 1EH, or if urgent, by telephone on 020 7242 9789 ext. 2620 and the committee administration will put you in contact with him.

Appendix 4. *Sample of Flier (DCD)*

Would you like to take part in a research project?

Children with Developmental Coordination Disorder (DCD)

(Researcher: Judith Peters)

Aim:

Most children easily learn to move about and use their hands and fingers for everyday activities. However there are some children who appear otherwise healthy and bright but have difficulty with daily activities e.g. learning to tie shoe-laces, fasten buttons, write neatly, ride a bike, hop or catch a ball. This study is being done to learn more about children (both with and without movement difficulties) so that we may be able to develop the best treatment programmes to help those children with movement problems

Who can take part?

- Boys aged from 7 to 10 years who attend mainstream school.
- The child should have a current diagnosis of Developmental Coordination Disorder (DCD). (often referred to as 'dyspraxia' in UK)
- The child should have no other illness or medical problem.
- The child should have normal hearing, and vision (with spectacles if necessary) and understand English.

How is the study being done?

If you are happy for your child to take part, your child will be invited to attend for a physiotherapy assessment at Great Ormond Street Hospital for Children. The assessment should involve a half-day session. Your child will be asked to do some every day activities, such as jumping, balancing like he/she does when playing with friends. Your child will also do some simple pencil and paper games. Your child is likely to find the activities fun and to enjoy the session.
Travel expenses would be covered for the visit to Great Ormond Street.

Who should I contact for further details?

Becky Worsley, Senior Physiotherapist
Children's Movement Project Coordinator
Physiotherapy Department, Level 5 Frontage Building
Great Ormond Street Hospital for Children
Great Ormond Street, London, WC1N 3JH
Tel: 020 7829 8610
e-mail: SmithR7@gosh.nhs.uk

Appendix 5. *Sample of Additional Information for Referrers (DCD)*

DCD Details for Referrals to GOSH Project (Judith Peters)

Developmental Coordination Disorder (DCD) 315.4 DSMIV

1. The child should meet all four criteria (A-D) listed below for a diagnosis of DCD:
   A. Performance in daily activities that require motor coordination is substantially below that expected given a person's chronological age and measured intelligence. This may be manifested by marked delays in achieving motor milestones e.g., walking, crawling, sitting), dropping things, "clumsiness", poor performance in sports, or poor handwriting.
   B. The disturbance in Criterion A significantly interferes with academic achievement or activities of daily living.
   C. The disturbance is not due to a general medical condition (e.g. cerebral palsy, hemiplegia, or muscular dystrophy) and does not meet the criteria for Pervasive Developmental Disorder.
   D. If mental retardation is present, the motor difficulties are in excess of those usually associated with it.

2. Interpretation for the present project:
   Criterion A
   The child should have failed a standardised movement test e.g. the Movement ABC or the Bruininks (score below 15th percentile). Definitely below average movement performance on a screening checklist (e.g. Movement ABC or DCDQ + clinical observation of movement difficulty would be acceptable for referral).

   Criterion B

   Parent/carers and or teachers should have expressed concern that the problem interferes significantly with daily activity/progress in school.

   Criterion C

   No medical condition other than 'DCD' to account for the movement difficulty. Does not better fit into Asperger's, Tourette's, or Attention Deficit Hyperactivity Disorder (ADHD) category.

   Criterion D

   For this project we intend to include children of average or above average intelligence who attend main steam school. (We shall confirm this with a short screening test).

3. Additional notes:
   The children should not have received a regular (significant) physiotherapy nor an occupational therapy movement programme within the last year. Ideally the children might come from a waiting list.

Initially we plan to recruit only **boys aged 7-10 years** however if we have insufficient participants we shall extend the age range and include girls.
Judith Peters will be 'blind' to the child's diagnosis. Please contact:
Becky Worsley, Senior Physiotherapist SmithR7@gosh.nhs.uk with any questions.

Thank you for your interest and help in this project Judith Peters, May 2003

Appendix 6. *Consent Form for Parents/Carers*

REC No. 03NS02 Version 1, dated 27/03/2003

Great Ormond Street Hospital for Children NHS Trust and Institute of Child Health Research Ethics Committee

Consent Form for PARENTS OR GUARDIANS of Children Participating in Research Studies

Title: Developmental Coordination Disorder (DCD): One syndrome with many subtypes, or one 'symptom' of may diffferent syndromes?

NOTES FOR PARENTS OR GUARDIANS

1. Your child has been asked to take part in a research study. The person organising that study is responsible for explaining the project to you before you give consent.

2. Please ask the researcher any questions you may have about this project, before you decide whether you wish to participate.

3. If you decide, now or at any other stage, that you do not wish your child to participate in the research project, that is entirely your right, and if your child is a patient it will not in any way prejudice any present or future treatment.

4. You will be given an information sheet which describes the research project. This information sheet is for you to keep and refer to. *Please read it carefully.*

5. If you have any complaints about the way in which this research project has been or is being conducted, please, in the first instance, discuss them with the researcher. If the problems are not resolved, or you wish to comment in any other way, please contact the Chairman of the Research Ethics Committee, by post via The Research and Development Office, Institute of Child Health, 30 Guilford Street, London WC1N 1EH or if urgent, by telephone on 020 7905 2620 and the committee administration will put you in contact with him.

CONSENT

I/We _____, being the parent(s)/guardian(s) of _____ agree that the Research Project named above has been explained to me to my/our satisfaction, and I/We give permission for our child to take part in this study. I/We have read both the notes written above and the Information Sheet provided, and understand what the research study involves.

SIGNED (Parent (s)/Guardian (s))	PRINTED	DATE
SIGNED (Researcher)	PRINTED	DATE

REC No. 03NS02 Version 1, dated 27/03/2003

Appendix 7. *Developmental History Questionnaire*

Child's Development and Contact Details for Project

It would be most helpful if you would provide some details about your child

Child's Details
Name: _____ Forename: _____

Gender: Male Female

Date of Birth (Day, Month, Year) _____

Family Contacts
Father's name: _____ Occupation: _____

Mother's name: _____ Occupation: _____

Address:

_____

_____

Postcode: _____

Telephone number: _____

School (Please note that the school will not be contacted without your permission)
Name of school: _____

Address_____

_____

Telephone number: _____

Is this a Mainstream School? Yes/No Is this a Special School? Yes/No

SEN Code of Practice:

School action: Does child receive extra help from school staff?	Yes	No
School action Plus: Does child receive help from external specialists?	Yes	No
Does child have a *Statement* of special educational need?	Yes	No
Is child being assessed for a *Statement* of special educational need?	Yes	No

Please put a ring round any words in the list below which refer to areas of difficulty for your child at school.
Reading Writing Attention Behaviour Catching a ball Running Falling over Balancing Doing up buttons Tying shoe laces Feeding Cutting with scissors

Has your child had physiotherapy or occupational therapy in the last 6 months? Y/N

Birth History
When was your child born? (weeks) 38-42 { } 33-37 { } 28-32 { } Under 28 { }

Birth weight _____

Normal delivery? Y/N. Caesarian? Y/N. Special Care Baby Unit needed? Y/N.

Developmental Milestones
Please give age at which child:

Sat ___ Walked _____ First word ____ Used sentences ____

Did your child crawl? Yes. No.

Please circle Child's hand used for writing: Right. Left. Ambidextrous (either).

Is mother father or any sibling left handed? Yes/No Ambidextrous? Yes/No

Medical History
Please write below any diagnosis your child has been given:

_____

_____

Please give details of any major illness or operation that your child has had:

_____

_____

_____

Please name any medication that your child is currently taking:

_____

_____

Vision and hearing
Vision: Normal? Y/N Corrected by spectacles? Y/N Treated for squint? Y/N

Hearing: Normal? Y/N History of hearing loss? Y/N Treated with grommits? Y/N

It would be helpful if you would give your child's ethnic origin?

_____

Signature of parent/carer who completed this form: _____

Please print name: _____

Date: _____

Thank you for providing us with this background information

Appendix 8. *Main Problem Area Questionnaire*

We should like you to tell us if your child has difficulty in any of the following areas: speech and language; concentration; overactivity; movement & coordination; relationships and friendships; reading; obsessive interests & routines; angry outbursts.

Please place these in order, from 1 (most problem area) to 8 (least problem area).

Start by marking the area you think causes most difficulty. Give this a score of 1.

Give the next area of concern a 2 and so on until you mark 8 for the area that causes least difficulty. Mark 0 if there is no problem in the area.

Area of Concern

Speech & language ☐

Concentration ☐

Overactivity ☐

Movement & coordination ☐

Relationships & friendships ☐

Reading ☐

Obsessive interests & routines ☐

Angry outbursts ☐

Child's name/ID: _____ Date: _____

Thank you for taking the time to complete this questionnaire

Appendix 9. *Coordination Flexibility Questionnaire*

Child's Name ___ Male/Female ____ Date of Birth _____

For each statement please put a tick in the box that you think is most like your child. It would help us if you put a tick for all the statements.

	Not True	Somewhat True	Certainly True
Looks awkward when running, hopping, or skipping			
Has difficulty writing			
Has dislocated a joint (put it out of place)			
Has difficulty dressing/undressing (tying shoe laces, fastening buttons etc)			
Has broken or fractured a bone			
Has sprained a joint more than once			
Has difficulty using tools such as cutlery, scissors			
Often trips up, bumps into people or things			
Muscles and joints are very flexible, mobile (seem 'double jointed')			
Often complains of aches and pains in limbs			

Appendix 10. *Strengths and Difficulties Questionnaire (Goodman, 1997)*

Strengths and Difficulties Questionnaire

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of the child's behaviour over the last six months or this school year.

Child's Name ..... Male/Female

Date of Birth .....

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	☐	☐	☐
Restless, overactive, cannot stay still for long	☐	☐	☐
Often complains of headaches, stomach-aches or sickness	☐	☐	☐
Shares readily with other children (treats, toys, pencils etc.)	☐	☐	☐
Often has temper tantrums or hot tempers	☐	☐	☐
Rather solitary, tends to play alone	☐	☐	☐
Generally obedient, usually does what adults request	☐	☐	☐
Many worries, often seems worried	☐	☐	☐
Helpful if someone is hurt, upset or feeling ill	☐	☐	☐
Constantly fidgeting or squirming	☐	☐	☐
Has at least one good friend	☐	☐	☐
Often fights with other children or bullies them	☐	☐	☐
Often unhappy, down-hearted or tearful	☐	☐	☐
Generally liked by other children	☐	☐	☐
Easily distracted, concentration wanders	☐	☐	☐
Nervous or clingy in new situations, easily loses confidence	☐	☐	☐
Kind to younger children	☐	☐	☐
Often lies or cheats	☐	☐	☐
Picked on or bullied by other children	☐	☐	☐
Often volunteers to help others (parents, teachers, other children)	☐	☐	☐
Thinks things out before acting	☐	☐	☐
Steals from home, school or elsewhere	☐	☐	☐
Gets on better with adults than with other children	☐	☐	☐
Many fears, easily scared	☐	☐	☐
Sees tasks through to the end, good attention span	☐	☐	☐

Signature ..... Date.....

Parent/Teacher/Other (please specify:)

Thank you very much for your help

Appendix 11. *High Functioning Autism Spectrum Screening Questionnaire (ASSQ) (Ehlers et al., 1999)*

Name of child ..... Date of birth .....

Name of rater ..... Date of rating .....

This child stands out as different from other children of his/her age in the following way:

	No	Somewhat	Yes
1. is old-fashioned or precocious	[ ]	[ ]	[ ]
2. is regarded as an "eccentric professor" by the other children	[ ]	[ ]	[ ]
3. lives somewhat in a world of his/her own with restricted idiosyncratic intellectual interests	[ ]	[ ]	[ ]
4. accumulates facts on certain subjects (good rote memory) but does not really understand the meaning	[ ]	[ ]	[ ]
5. has a literal understanding of ambiguous and metaphorical language	[ ]	[ ]	[ ]
6. has a deviant style of communication with a formal, fussy, old-fashioned or "robotlike" language	[ ]	[ ]	[ ]
7. invents idiosyncratic words and expressions	[ ]	[ ]	[ ]
8. has a different voice or speech	[ ]	[ ]	[ ]
9. expresses sounds involuntarily; clears throat, grunts, smacks, cries or screams	[ ]	[ ]	[ ]
10. is surprisingly good at some things and surprisingly poor at others	[ ]	[ ]	[ ]
11. uses language freely but fails to make adjustment to fit social contexts or the needs of different listeners	[ ]	[ ]	[ ]
12. lacks empathy	[ ]	[ ]	[ ]
13. makes naive and embarrassing remarks	[ ]	[ ]	[ ]
14. has a deviant style of gaze	[ ]	[ ]	[ ]
15. wishes to be sociable but fails to make relationships with peers	[ ]	[ ]	[ ]
16. can be with other children but only on his/her terms	[ ]	[ ]	[ ]
17. lacks best friend	[ ]	[ ]	[ ]
18. lacks common sense	[ ]	[ ]	[ ]
19. is poor at games: no idea of cooperating in a team, scores "own goals"	[ ]	[ ]	[ ]
20. has clumsy, ill coordinated, ungainly, awkward movements or gestures	[ ]	[ ]	[ ]
21. has involuntary face or body movements	[ ]	[ ]	[ ]
22. has difficulties in completing simple daily activities because of compulsory repetition of certain actions or thoughts	[ ]	[ ]	[ ]
23. has special routines: insists on no change	[ ]	[ ]	[ ]
24. shows idiosyncratic attachment to objects	[ ]	[ ]	[ ]
25. is bullied by other children	[ ]	[ ]	[ ]
26. has markedly unusual facial expression	[ ]	[ ]	[ ]
27. has markedly unusual posture	[ ]	[ ]	[ ]

Specify reasons other than above:

Appendix 12. *Score Sheet for Experimental Tests*

ID: _____ **Date:** _____

Counting in 3s good average poor **Counting in 2s** good average poor

Peg board Preferred hand R L

Turning 16 small pegs **Preferred** _____ **Non Preferred** _____

Turning 16 small pegs **Preferred** _____

While counting forward in 3s

Placing 16 pegs

(8 large 8 small) **Preferred** _____ **Non Preferred** _____

O O O O O O O O

O O O O O O O O

O O O O O O O O

O O O O O O O O

Balance Preferred leg R L

Balance on one leg **Preferred** _____ **Non-preferred** _____
up to 30 secs (eyes open)

Balance on one leg **Preferred** _____ **Non-preferred** _____

While counting in 3s

Balance on one leg **Preferred** _____ **Non-preferred** _____
up to 30 secs (eyes closed)

Comment:

Appendices 13 a-e ***Individual data for final groups Study 5 Chapter 9***

Appendix 13a. *Developmental Coordination Disorder Final Group (DCDf) individual data*

ID	Uneven peg place preferred hand	Small peg turn preferred hand	Small peg turn preferred hand (+ counting)	Small peg tun/counting difference	Balance preferred leg (vision)	Balance preferred leg (no	Balance preferred leg vision/no vision difference	Balance preferred leg (vision +counting)	Balance preferred leg/counting difference
9	42.3	50.6	44.8	5.8	3.5	3.0	.5	5	-1.5
16	36.2	36.2	34.9	1.3	30.0	18.2	11.8	30	.00
24	42.5	47.2	69.0	-21.8	2.1	1.0	1.1	25.6	-23.5
3	61.8	132.4	75.3	57.1	3.9	4.6	-.7	7.9	-4.0
4	78.0	72.3	68.3	4.0	4.9	1.4	3.5	10.3	-5.4
6	31.8	31.1	30.8	.3	22.0	5.2	16.8	16.6	5.4
7	47.3	32.1	62.6	-30.5	30.0	8.4	21.6	5.7	24.3
8	37.3	36.7	37.8	-1.1	13.2	6.5	6.7	20	-6.8
13	51.7	49.5	68.3	-18.8	15.4	1.0	14.4	11.9	3.5
15	50.6	38.5	69.4	-30.9	9.0	1.3	7.7	17.8	-8.8
17	42.8	34.9	30.4	4.5	30.0	2.0	28.0	N/A	N/A
20	44.3	41.7	43.9	-2.2	30.0	3.8	26.2	13.9	15.1
25	45.3	42.4	37.4	5.0	30.0	2.4	27.6	30.0	-2.2

Appendix 13b. ***High Functioning Autism Final Group (HFAf) Individual Data***

ID	Uneven peg place preferred hand	Small peg turn preferred hand	Small peg turn preferred hand (+ counting)	Small peg tun difference	Balance preferred leg (vision)	Balance preferred leg (no vision)	Balance difference	Balance preferred leg (vision +counting)	Balance preferred leg/counting difference
2	N/A	44.9	N/A	N/A	N/A	N/A	N/A	N/A	N/A
11	35.0	38.9	44.3	-5.4	30.0	6.1	23.9	30.0	.00
18	61.0	42.7	60.0	-17.3	30.0	7.5	22.5	17.3	12.7
21	56.3	74.0	67.7	6.3	5.0	2.0	3.0	8.2	-3.2
35	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
28	74.4	62.0	51.4	10.6	N/A	N/A	N/A	N/A	N/A
29	52.8	53.0	N/A	N/A	N/A	N/A	N/A	N/A	N/A
30	63.5	64.5	42.3	22.2	9.8	3.0	6.8	6.0	3.8
32	37.8	35.2	41.9	-6.7	30.0	2.5	27.5	10.6	19.4
36	35.0	48.6	46.3	2.3	16.3	8.3	8.0	20.2	-3.9
38	61.8	44.8	N/A	N/A	4.0	2.0	2.0	3.2	.8
39	72.2	70.0	101.2	-31.2	14.6	2.0	12.6	8.1	6.5

Appendix 13c. ***Typically Developing Final Group (TDf) Individual Data***

ID	Uneven peg place preferred hand	Small peg turn preferred hand	Small peg turn preferred hand (+ counting)	Small peg tun difference	Balance preferred leg (vision)	Balance preferred leg (no vision)	Balance difference	Balance preferred leg (vision +counting)	Balance preferred leg/counting difference
40	47.2	49.1	51.1	-2.0	30.0	20.3	9.7	30.0	.00
41	36.7	31.1	46.7	-15.6	30.0	3.8	26.2	30.0	.00
42	32.8	28.4	24.1	4.3	30.0	8.0	22.0	30.0	.00
43	46.5	33.8	33.1	.7	28.8	2.2	26.6	30.0	-1.2
44	48.7	41.9	79.3	-37.4	30.0	10.5	19.5	30.0	.00
46	42.9	42.1	42.4	-.3	30.0	18.6	11.4	30.0	.00
47	38.7	41.6	96.8	-55.2	26.8	10.7	16.1	30.0	-3.2
48	60.4	40.5	72.1	-31.6	N/A	N/A	N/A	N/A	N/A
49	29.6	39.0	29.9	9.1	30.0	7.3	22.7	21.8	8.2
50	38.6	36.8	37.7	-.9	30.0	6.0	24.0	30.0	.00

Appendix 13d. ***Motor Difficulty Hypermobile Group (MD-hyp) Individual Data***

ID	Uneven peg place preferred hand	Small peg turn preferred hand	Small peg turn preferred hand (+ counting)	Small peg tun difference	Balance preferred leg (vision)	Balance preferred leg (no vision)	Balance difference	Balance preferred leg (vision +counting)	Balance preferred leg/counting difference
9	42.3	50.6	44.8	5.8	3.5	3.0	.5	5.0	-1.5
16	36.2	36.2	34.9	1.3	30.0	18.2	11.8	30.0	.00
24	42.5	47.2	69	-21.8	2.1	1.0	1.1	25.6	-23.5
2	N/A	44.9	N/A	N/A	N/A	N/A	N/A	N/A	N/A
11	35.0	38.9	44.3	-5.4	30.0	6.1	23.9	30.0	.00
18	61.0	42.7	60.0	-17.3	30.0	7.5	22.5	17.3	12.7
21	56.3	74.0	67.7	6.3	5.0	2.0	3.0	8.2	-3.2
35	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
27	46.2	45.4	45.9	-.5	20.5	2.0	18.5	16.4	4.1
45	33.7	29.9	28.5	1.4	30.0	14.8	15.2	30.0	.00
14	57.3	43.3	99.7	-56.4	16.4	1.0	15.4	5.2	11.2

Appendix 13e. ***Motor Difficulty Non-hypermobile (MD-nonhyp) Group Individual Data***

ID	Uneven peg place preferred hand	Small peg turn preferred hand	Small peg turn preferred hand (+ counting)	Small peg tun difference	Balance preferred leg (vision)	Balance preferred leg (no vision)	Balance difference	Balance preferred leg (vision +counting)	Balance preferred leg/counting
3	61.8	132.4	75.3	57.1	3.9	4.6	-.7	7.9	-4.0
4	78.0	72.3	68.3	4.0	4.9	1.4	3.5	10.3	-5.4
6	31.8	31.1	30.8	.3	22.0	5.2	16.8	16.6	5.4
7	47.3	32.1	62.6	-30.5	30.0	8.4	21.6	5.7	24.3
8	37.3	36.7	37.8	-1.1	13.2	6.5	6.7	20.0	-6.8
13	51.7	49.5	68.3	-18.8	15.4	1.0	14.4	11.9	3.5
15	50.6	38.5	69.4	-30.9	9.0	1.3	7.7	17.8	-8.8
17	42.8	34.9	30.4	4.5	30.0	2.0	28.0	N/A	N/A
20	44.3	41.7	43.9	-2.2	30.0	3.8	26.2	14.9	15.1
25	45.3	42.4	37.4	5	30.0	2.4	27.6	30.0	-2.2
28	74.4	62	51.4	10.6	N/A	N/A	N/A	N/A	N/A
29	52.8	53.0	N/A	N/A	N/A	N/A	N/A	N/A	N/A
30	63.5	64.5	42.3	22.2	9.8	3.0	6.8	6.0	3.8
32	37.8	35.2	41.9	-6.7	30.0	2.5	27.5	10.6	19.4
36	35.0	48.6	46.3	2.3	16.3	8.3	8.0	20.2	-3.9
38	61.8	44.8	N/A	N/A	4.0	2.0	2.0	3.2	.8
39	72.2	70.0	101.2	-31.2	14.6	2.0	12.6	8.1	6.5
31	N/A	72.5	53.2	19.3	21.3	3.8	17.5	30.0	-8.7
51	52.1	59.2	63.0	-3.8	23.1	13.4	9.7	15.6	7.5
12	29.3	27.1	32.8	-5.7	30.0	7.1	22.8	7.0	23.0

Appendix 14 *Questionnaire used in Study 2 Chapter 6*

Physiotherapy Department Survey

The Physiotherapy Department GOSH for Children NHS Trust is carrying out a survey about the delivery of physiotherapy services to children seen in the department.

We would be grateful if you would spare a few minutes to complete the questionnaire. A reply envelope is enclosed.

How to fill in this form:
Please tick the appropriate box and write in the spaces provided for comments.
If you cannot or do not wish to answer a particular question, then please leave the question blank. **All responses will be treated in the strictest confidence.**

Questions:

1. Please tick any of the labels/diagnosis given to your child. Place a * by the label that you use / prefer.

- ☐ Dyspraxia
- ☐ Developmental Co-ordination Disorder (DCD)
- ☐ Attention Deficit Disorder (ADD)
- ☐ Hyperactive
- ☐ Attention Deficit Hyperactivity Disorder (ADHD)
- ☐ Clumsy
- ☐ Incoordination Difficulty
- ☐ Sensory Integrative Dysfunction / disorder
- ☐ Motor Learning Difficulty
- ☐ Other (please specify) _____

2. What was the main problem area that lead to a physiotherapy referral?

- ☐ Gross Motor Function
- ☐ Fine Motor Function
- ☐ Attention
- ☐ Organisation
- ☐ Handwriting
- ☐ Don't know
- ☐ Other (please specify) _____

3. Who suggested that your child should be referred for physiotherapy?

- ☐ GP
- ☐ Doctor within GOSH
- ☐ Other doctor outside GOSH
- ☐ Physiotherapist
- ☐ Teacher
- ☐ Psychologist
- ☐ Child's parent/carer
- ☐ Other ....(please specify) _____
- ☐ Don't know.

4. How long after referral did you wait to receive an initial physiotherapy appointment at GOSH?

- [ ] Less than 1 month
- [ ] 1 - 2 months
- [ ] 3 - 4 months
- [ ] 5 - 6 months
- [ ] 7 - 8 months

Over 9 months (please specify) _____

5. Please rate the various aspects of the physiotherapy appointment by making
   0 = Unsatisfactory 1 = Satisfactory

- [ ] Time waiting for appointment
- [ ] Convenience of appointment
- [ ] Information about what to expect of initial appointment
- [ ] Information about how to find the physiotherapy department.
- [ ] Other (please specify) _____

6. Did you feel that the physiotherapist identified your child's difficulties ?

- [ ] Very well
- [ ] Quite well
- [ ] Unsure
- [ ] Not very well
- [ ] Not at all

7. Was your child offered physiotherapy treatment sessions at GOSH ?

- [ ] Yes
- [ ] No

8. If yes. How many physiotherapy sessions did your child attend?

- [ ] Declined to attend session
- [ ] One session
- [ ] 2 - 6 sessions
- [ ] 7 - 11 sessions
- [ ] 12 or more sessions.

9. Please rate the value of the treatment provided ?

- [ ] Very unhelpful
- [ ] Unhelpful
- [ ] Unsure
- [ ] Helpful
- [ ] Very helpful

10. Did the physiotherapist provide advice / home programme?

- [ ] Yes
- [ ] No

11. Please rate the advice/home programme provided ?

- [ ] Very unhelpful
- [ ] Unhelpful
- [ ] Unsure
- [ ] Helpful
- [ ] Very helpful

12. Did you find the physiotherapy programme ?

- [ ] Very clear and easy to follow
- [ ] Fairly clear and easy to follow
- [ ] Unsure
- [ ] Rather unclear and hard to follow
- [ ] Very unclear and hard to follow

13. How often did/does your child carry out the physiotherapy programme?

- [ ] Every day
- [ ] 1 day a week
- [ ] 2 - 6 days a week
- [ ] Other - (please specify) _____
- [ ] Never

14. Please rate the various aspects of the physiotherapy service by marking
0 = not helpful 1 = helpful 2= very helpful

- [ ] Observing assessment
- [ ] Home programme
- [ ] Physiotherapy report
- [ ] Physiotherapy treatment sessions
- [ ] Handwriting advice
- [ ] Opportunity to talk about child's difficulty
- [ ] Advice for school
- [ ] Advice for leisure activity
- [ ] Information and leaflets
- [ ] Other - (please specify) _____

15. What effect do you feel that physiotherapy had on your child's condition?

- [ ] Very much improved
- [ ] Improved
- [ ] No change
- [ ] Worse
- [ ] Very much worse
- [ ] Changed but not due to physiotherapy.

16. Who paid for your child's physiotherapy?

- [ ] Yourself
- [ ] NHS
- [ ] G.P fundholder
- [ ] BUPA or other private insurance
- [ ] Other (please specify) _____
- [ ] Don't know

17. How would you rate the physiotherapy service you received with regard to value for money?

- ☐ Very good value
- ☐ Good value
- ☐ Average
- ☐ Poor value
- ☐ Very poor value
- ☐ Unsure

Please comment on any aspect of the dyspraxia/DCD service and/or make suggestions as to how it may be improved.

Comments:

Thank you very much for completing this questionnaire.

Please return it in the envelope provided to:
Devala Dookun MCSP (Ms)
Head of Physiotherapy

Please feel free to add a comment about your child's present progress/difficulty. If you feel that a further physiotherapy review of your child is required please leave your name, and telephone number and your child's name and date of birth and we will contact you.